The Journal of
Bone and Joint Surgery

EDITORIALS AND ANNOTATIONS

JOINT MEETING OF THE ORTHOPAEDIC ASSOCIATIONS OF THE ENGLISH-SPEAKING WORLD—MAY 1958

It has happened again! It has been happening since our orthopaedic forefathers, John Ridlon and Robert Jones, crossed the Atlantic Ocean by steam packet to discover what was known of the treatment of diseases of joints on the other side. It happened when Goldthwait brought his team of surgeons to help Great Britain in its hours of travail early in the first world war, and when our forces were mingled during the second war. It happened when the American and British Orthopaedic Associations first met together in London in 1929, when both were the guests of the newly formed Canadian Association in Quebec in 1948, and when all three were joined by the nations of the British Commonwealth in London in 1952. It happens each year when the Travelling Fellows, the flower of a rising generation, are warmed in the sun of another country. But it is the same sun that warms the whole world; and humanity, like the sun, knows no boundaries: of course it has happened again!

This, the second occasion on which the Orthopaedic Associations of the English-speaking world have joined in a combined meeting, has been no less memorable than the first. Indeed, a meeting held in the Capital City of the United States of America, and under the auspices of such hosts as the American Orthopaedic Association, could hardly be otherwise. A great number of surgeons were attracted, many travelling across a continent or an ocean, a few half way around the world. It was sad that the New Zealand representatives were prevented at the last minute from making the journey, but eight men came from Australia, and twelve from South Africa. The Canadian Association was present in great strength, and from the United Kingdom there were about one hundred. The total number registered, including ladies, was no less than 992. That so many were able to cross the Atlantic from Great Britain was due in no small measure to the generosity of the Washington Orthopaedic Club, who gave hospitality to forty of them in their homes, and to many more in an hotel. The British Orthopaedic Association presented the Club with a " loving cup " as a token of its appreciation of a magnificent gesture that made it possible for many of its younger members and associates to attend the meeting.

The scientific programmes had been most carefully arranged; they included contributions from members of all the participating Associations and achieved a very high standard. The formal sessions occupied half of each day; in the other half, scientific demonstrations and an attractive audio-visual programme competed with a variety of social activities. Two of the formal sessions were held in the auditoria of the National Institutes of Health and the Walter Reed Army Medical Centre, and these were followed by demonstrations and tours arranged by these institutions. There were also tours of the Armed Forces Institute, the National Naval Hospital and the Washington Hospital Centre. These visits did make clear the immense resources that are being devoted by the United States to research. The annual expenditure of the National Institutes of Health must alone be greater than the combined budgets of every department for medical research in the whole of the United Kingdom. Visitors are inevitably filled with awe at the scale of these projects and cannot help wondering how they themselves, with their puny resources, ever manage to make a worthwhile contribution.

This is not the place to elaborate on the scientific proceedings. They will be reported in abstract in the July issue of the American Volume, and many of the papers will be published

later in either the American or the British Volume. An account of the wonderful tour through the Eastern States and Canada that was arranged after the meeting is given in the Proceedings Section of this issue. We are concerned here with an even more important matter—the meeting of people, people from different parts of the world, and of varying habits, but having a common language, a common culture, and common ideals. Nothing but good can come from such a meeting. This was fully appreciated by the President of the United States, who graciously received a few representatives from each country. The occasion was informal, and he made it clear that it was his view that the significance of an international congress extends far beyond the mere interchange of scientific knowledge. His words are quoted in full elsewhere in this number. In a gathering of this size it could easily happen that there were few opportunities for intimate contact, but the social activities had been so skilfully arranged that the reverse was the case. There was such a wide choice that the groups were comparatively small, and these quickly split up again. Small groups of men and women were always to be seen in discussion, and, from the snatches of conversation overheard, it was clear that they were not merely exchanging social formalities, or even arguing medical topics; the talk ranged over the whole field of the social and political problems of their different countries.

The ladies quickly became acquainted in the Hospitality Lounge and at a number of sight-seeing trips—a tour of the National Gallery of Art, where the new Salvador Dali is said to have attracted more attention than the old masters; visits to the Smithsonian Institution, the Arlington Cemetery, the White House, the Houses of Congress and the Federal Bureau of Investigation; and, by way of contrast, entertainment on the foreign soil of the British and New Zealand Embassies. Will any one of them ever forget driving through Maryland in spring with the azalias in full bloom? Which of them could fail to be impressed by the voyage down the Potomac river to the Colonial home of George Washington? And when they were joined by their men-folk at the Reception of the President of the American Orthopaedic Association and at the Reception of the President of the Canadian Orthopaedic Association it seemed as if this great concourse of people were age-old friends. These two receptions were the highlights of the social activities; they were held on the beautiful lawn of the Shoreham Hotel on perfect cloudless evenings, and the sound of a thousand voices drifted away to the skies.

The A.B.C. Club owes its existence to the meeting in Quebec in 1948, yet another mark to the credit of joint meetings. It is hard to say who was first responsible for organising the travelling fellowships, but surely most of the credit must go to the then Presidents of the British and American Associations, Alan Malkin and Bob Harris. The fellowships have now become a much valued institution and, as always, the Travelling Fellows were a prominent feature of the meeting. This time the travellers were British, and Martin Brett, Bob Bremner, Robert Owen and Noel Blockey from the United Kingdom, and Manuel Lanz from South Africa, were present as the guests of the American Orthopaedic Association. It is interesting to look over the list of members of the A.B.C. Club—they number forty-eight excluding the five travellers of this year—and see if those of us responsible for their selection carried out our task wisely. The British visitors to America in 1948 numbered thirteen, in 1954 there were four from the United Kingdom and one Australian, and in 1956 four from the United Kingdom and one New Zealander. Ten from the United States and five from Canada visited Britain in 1949, and four from the United States and one Canadian in each of the years 1955 and 1957. There is, of course, no real yardstick by which the worth of a surgeon can be measured, but every single one of these has made or is rapidly making his mark. Election to Membership of the American Orthopaedic Association, and to Fellowship of the British Orthopaedic Association, gives some indication of how they are judged by their peers. Of the original thirteen from the United Kingdom, eleven are now Fellows, and every one of the first ten from the United States are now Members (the latter perhaps because they survived the ordeal by "deep-freeze"!). Of the 1954–55 vintage, two from the United States and two British have been elected, and of the 1956–57 travellers, one British has already achieved this

distinction. The A.B.C. Club are indeed becoming a force in orthopaedics, and this link between America, Britain and all the British Commonwealth should prove a powerful bond.

The history of these combined meetings is entirely good. There can be no doubt of their value from the scientific, the social, and most particularly the international points of view. The news that yet another is being planned for 1964 is therefore most welcome. Although it is not yet certain in which country it is being held, it will surely prove to be a magnet that will draw together once more the Presidents' jewels of office, the proud symbol of the friendship that has always existed between the orthopaedic surgeons of the world. PHILIP WILES.

THE JOURNAL

The combined meeting of the Orthopaedic Associations provided the opportunity for an important meeting of the Board of Trustees and the British Editorial Board. The agreement between the two bodies calls for an annual meeting of their representatives, but it is seldom that more than one or two can be present. On the occasion of a combined meeting, however, a much larger attendance is possible: those at Quebec in 1948 and London in 1952 were fully representative, and much useful business was transacted. This time the two Boards met in Baltimore as the guests of Dr R. W. Johnson, seven out of the eight members of the Board of Trustees and ten members of the British Board being present. Dr William Rogers and Sir Reginald Watson-Jones, who were kept away by illness, were both sadly missed. The British Editor, however, sent a recorded message that was played at the opening of the meeting.

The Journal of Bone and Joint Surgery has a bigger circulation and publishes a larger number of pages than any comparable specialist periodical. The work of the Editorial Boards in maintaining its high standard is arduous, and the managers, who are conducting a sizeable business, have an equally responsible task. Business methods on the two sides of the Atlantic vary considerably; there are differences in the details of office management, in the methods of production, and in such matters as Governmental rules controlling the conduct of charitable organisations. The difficulty of explaining these differences by correspondence is considerable, and it is almost impossible to convey the more delicate nuances of feeling that contribute so much to harmonious working. But when men of goodwill meet, difficulties are soon resolved. And the Journal is conducted by men of goodwill, men inspired by the same ideal—the service they can render to all humanity by making easily available the best that is known of the art and science of orthopaedic surgery. In ten years of co-operation there has been no discord in editorial policy. The problem has never been the end, only the means by which the end is achieved. Mundane matters could not be allowed to wreck so fine an enterprise, and once again the differences between the two Boards have been resolved. Co-operation in the production of the Journal is assured for a further term of years, and the new formula is so simple that there now seems to be no reason why it should not continue indefinitely.

The Journal, conceived and nurtured by the American Orthopaedic Association, was born at the end of the last century and named *Transactions*, but soon it became the *American Journal of Orthopaedic Surgery*. In 1919 the name was changed to the *Journal of Bone and Joint Surgery* and at the same time it became also the official publication of the British Orthopaedic Association. In 1936 the hospitality of its pages was extended to the American Academy of Orthopaedic Surgeons. Ten years ago the American Orthopaedic Association, with characteristic generosity, took the British Orthopaedic Association into partnership and the British Volume, representing the practice of the surgeons of the British Commonwealth, first appeared. The last stage in development was in 1954, when the Academy was given a share in the control of and financial responsibility for the American volume. Thus the child of the American Orthopaedic Association, reared with the energy and money of its members, and the hard work, almost the slavery of its Editors, has become in its years of maturity a supreme example of successful international co-operation. PHILIP WILES.

VERTEBRA PLANA

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Since 1925, when Calvé first described the condition that bears his name, many other cases have been reported. But in each case the emphasis has been on the immediate radiological appearance, the diagnosis or the treatment.

Fairbank (1952) was the first to suggest a cause other than osteochondritis with his statement: " Personally I feel that eventually verteba plana will be proved to be one manifestation of granulomatosis, and not a clinical entity "; and in 1954 Compere, Johnson

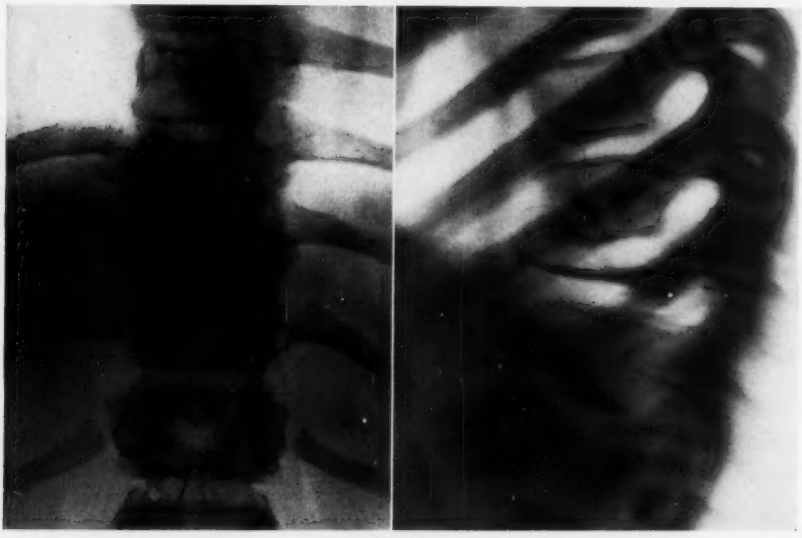

FIG. 1

Case 1—June 1936. Age nine years. One month after onset (Royal National Orthopaedic Hospital).

and Coventry reported four cases in which biopsy revealed eosinophilic granuloma. In their cases treatment consisted either of deep x-ray therapy or of a combination of deep x-ray and bone grafting. Although partial regeneration of the affected vertebra had occurred in one case, no case was followed long enough to determine the final clinical condition or radiological appearance.

At an informal meeting I reported Case 1 of this series, which had been followed for twenty-two years; colleagues present were able to report similar cases and have kindly allowed me to include them in the series.

Case 1—A girl born in 1927 first attended hospital in October 1935 with pain in the back which dated from a fall on to her back six weeks before. She had lost weight and suffered from night sweats.

On examination there was a slight mid-thoracic kyphosis with a minimal right thoracic and left lumbar scoliosis. Extension and flexion were limited and painful. Radiographs showed complete collapse of the tenth thoracic vertebra with increased density (Fig. 1).

She was admitted to the country section of the Royal National Orthopaedic Hospital at Stanmore and was kept recumbent on a frame until April 1936. The Mantoux test was negative in November 1935 but faintly positive in January 1936. In July 1937 she complained

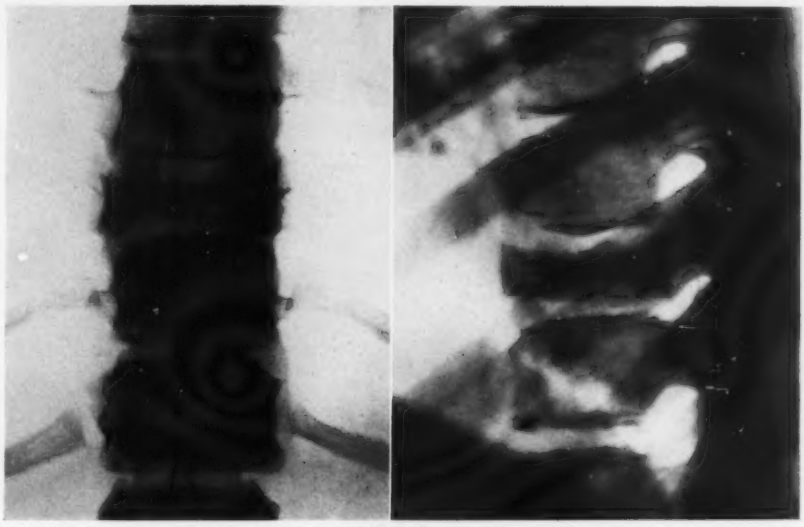

FIG. 2
Case 1—Three years after onset.

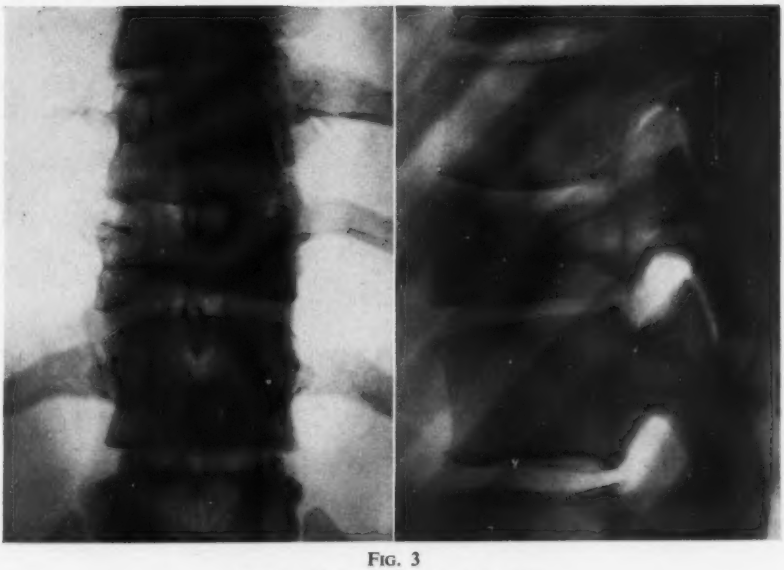

FIG. 3
Case 1—Twenty-two years after onset.

of a slight transient ache in the back, but had no other symptoms up to 1939, when because of the war she moved out of London and was not seen again until she was examined recently, twenty-two years after her first attendance. She is now married and has had three confinements.

FIG. 4
Case 2—October 1935. Age seven years. Eighteen months after onset (Royal National Orthopaedic Hospital).

Clinically there is no abnormality in her back and she is free from symptoms.

The radiographs of 1939 show an intermediate stage in the regeneration (Fig.2). Recent radiographs show a regeneration of the affected vertebra, the depth being about three-quarters of the normal. There is a line of increased density visible in the centre corresponding to the original condensed area (Fig. 3). A complete skeletal survey was not carried out.

Case 2 (Mr H. J. Seddon's case)—A boy born in 1928 first attended the Royal National Orthopaedic hospital in February 1935 at the age of seven and a half years, complaining of pain in the back. There had been a fall a few weeks before, but this does not appear to have been significant.

On examination there was a slight lower thoracic kyphosis with tenderness over the gibbus and some rigidity of the spine. Radiographs showed complete collapse of the ninth thoracic vertebra; the joint spaces above and below were normal (Fig. 4). The Mantoux and Wassermann tests were negative. He was treated on a frame until October 1935. He served in the Royal Navy in the second world war without disability or symptoms.

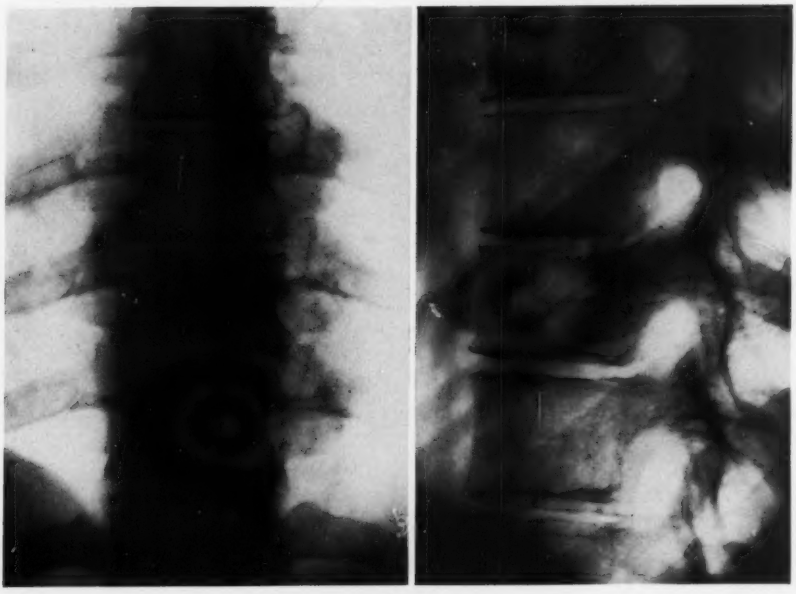

FIG. 5
Case 2—Twenty-two years later.

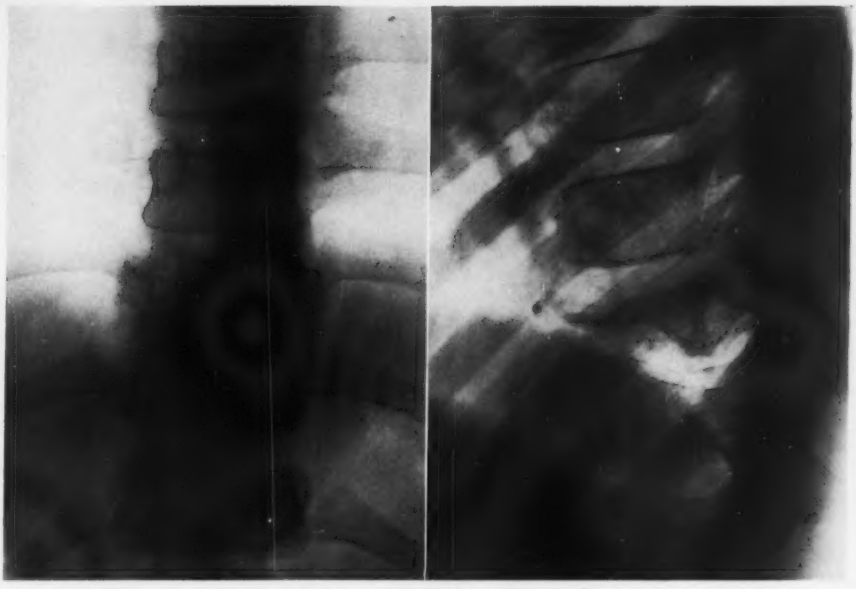

FIG. 6
Case 3—August 1945. Age twelve years. Four months after onset (Alexandra Hospital, Luton).

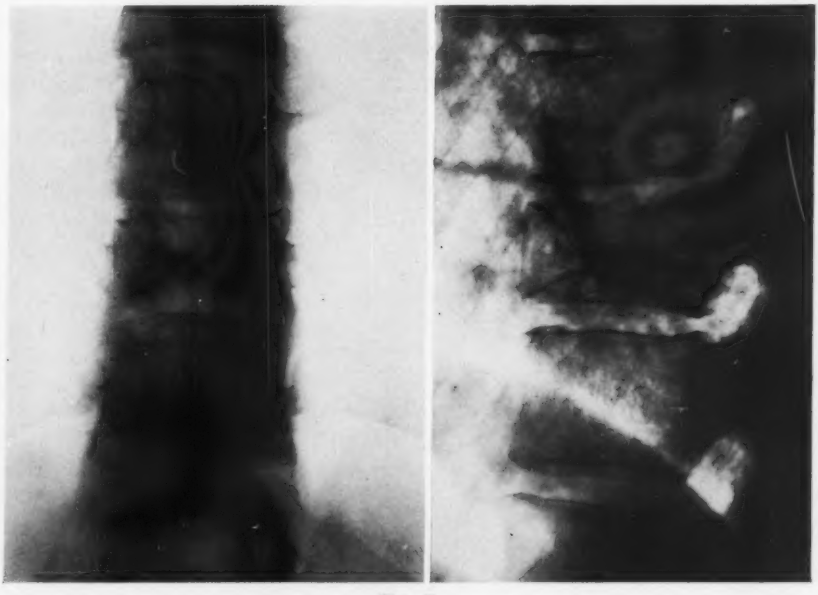

FIG. 7
Case 3—Twelve years after onset (St Bartholomew's Hospital, London).

In May 1957 he was re-examined, twenty-two years after his first attendance. He was free from symptoms, there was no tenderness in the back and no deformity. Radiographs showed that the vertebral body had regenerated and was now two-thirds of the normal height. There was no line of increased density in the middle of the vertebra (Fig. 5).

Case 3 (Mr W. D. Coltart's case)—A boy born in June 1933 first complained of pain in the middle of the back in April 1945. He was later admitted to the Alexandra Hospital, Luton. Radiographs showed complete collapse of the front of the tenth thoracic vertebra, but the back had retained about two-thirds of its normal height (Fig. 6). He was treated first by bed rest and from September 1945 in a plaster bed. In July 1946 he started to get up in a plaster jacket, which was continued for one month.

In April 1957, twelve years after his first attendance, he was examined at St Bartholomew's Hospital. He was free from symptoms, and clinically the back was normal. Radiographs showed regeneration of the vertebra with a line of increased density in the centre of the body (Fig. 7).

Case 4 (Mr H. Jackson Burrows's case)—A girl born in January 1932 fell on her back in April 1935 with her " identical " twin sister on top of her. Thereafter she would wake up at night screaming. The tuberculin test carried out next month was negative, but a radiograph (May 1935) showed flattening of the greater part of the body of the eleventh thoracic vertebra, which looked very dense (Fig. 8). She was treated in Queen Mary's Hospital, Carshalton, for two years (May 1935 to May 1937), at first on a frame and then in a celluloid jacket, in which she was discharged. Seen six months later (November 1937) at the age of five years and eleven months, she had no deformity, but some stiffness of the back ascribed to prolonged immobilisation. Serial radiographs showed progressive restoration of the vertebra. She was last seen and radiographed as a child in June 1939 at the age of seven and a half (Fig. 9). The outbreak of war found her hop-picking; the jacket broke and she discarded it, without any obvious ill-effect. She remained out of London because of the war. At the age of fourteen (1945) she began sedentary work, dressmaking; at seventeen (1948) she changed to standing work as an inspector. At twenty-one (1952) she married, and she has had two children, now aged three years and two years. Seen twenty-two years after her first attendance, at the age of twenty-five, she reported no trouble referable to her thoracic spine. For three years she has had a mild low lumbar ache after standing an hour or an hour and a half, especially stooping as in ironing, but she does not consider that this discomfort warrants treatment. Her back appears normal on clinical examination. Radiographs (Fig. 10) show very slight narrowing of the front of the body of the eleventh thoracic vertebra. A horizontal dense line is visible behind its centre.

DISCUSSION

These four patients came under treatment between 1935 and 1945, and no biopsy was performed; therefore no fresh light is thrown upon diagnosis, but sufficient time has passed to show the end-result.

The cases were typical in their mode of onset, symptomatology and clinical findings. The early radiographic appearances were also typical. The patients were all treated by a period of recumbency but with no deep x-ray therapy, and the recent follow-up shows that in each case there was a complete relief of symptoms without any appreciable clinical abnormality. Only in Case 2 has a complete skeletal survey been carried out, and it shows no other bone lesion. In each case later radiographs show almost complete regeneration of the affected vertebra and in three of the cases there is a narrow line of sclerosis (the ghost vertebra) representing the original collapsed and dense vertebral body.

Clearly, a skeletal survey would now be carried out on any patient presenting with similar radiographic appearances, but it appears that solitary lesions of this type in the

FIG. 8

FIG. 9

Case 4. Figure 8—May 1935. Age three years and five months. One month after onset (Queen Mary's Hospital, Carshalton). Figure 9—Four years later. Age seven years and six months (Miller General Hospital, Greenwich).

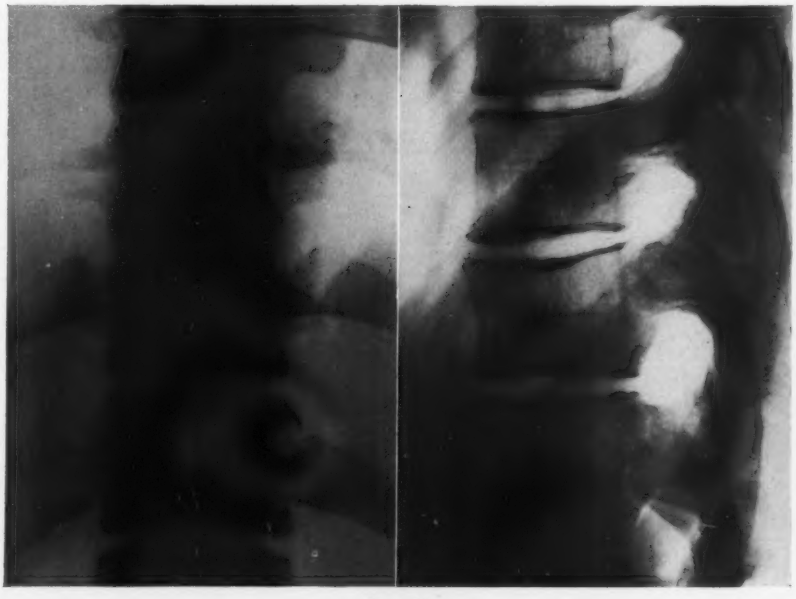

FIG. 10

Case 4—Twenty-two years after onset. Age twenty-five (Royal National Orthopaedic Hospital).

vertebral column can be self-limiting. A review of these patients suggests that in solitary lesions deep x-ray treatment is not necessary, provided the patient can be kept under supervision with repeated radiological examination in order that the process of regeneration can be followed. In view of the possible after-effects of deep x-ray treatment it is desirable to limit this to cases in which it is proved to be necessary.

If there is radiological evidence of multiple lesions, a biopsy should be performed on one that is accessible, and appropriate deep x-ray treatment instituted. There is no suggestion of withholding treatment in this group of cases.

I wish to express my thanks to Mr H. Jackson Burrows, Mr W. D. Coltart and Mr H. J. Seddon for allowing me to include their patients in my series, and to the Medical Records Department of the Royal National Orthopaedic Hospital for tracing two of the patients who had married and moved away from London.

Figures 8 and 9 are from prints, discoloured and faded from subjection to heat and water, salvaged from the premises of Messrs John Wright & Sons which were destroyed in a night bombardment of Bristol.

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DIAPHYSO-EPIPHYSIAL RESECTION FOR BONE TUMOUR AT THE KNEE

With Reports of Nine Cases

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Extensive diaphyso-epiphysial resection is advisable for bone tumours when amputation appears too drastic and local excision not sufficient.

Indications for resection

Resection is to be contemplated in the following cases.

Giant-cell tumours may demand resection in two circumstances: 1) recurrence after curettage once or twice, supplemented or not by radiotherapy; 2) as the primary treatment when bone destruction does not allow complete excision, or when clinical and radiological features, together with histological findings, show a probability of malignant evolution.

Certain malignant tumours (chondrosarcoma, fibrosarcoma, periosteal sarcoma) show a very slow evolution, making a decision about amputation particularly difficult and questionable.

Even in *osteogenic sarcoma*, amputation may be refused by the patient or his family, and may not always be justified by a much higher rate of survival.

Restoration of continuity after resection

For tumours at the knee (lower end of the femur and upper end of the tibia) resection is often rejected on account of the technical difficulties of restoring continuity of the bone.

Among the many procedures that have been advocated, those that try to preserve the mobility of the joint either by a prosthesis or by a massive homogenous graft still appear quite unreliable. Prostheses usually loosen or break after a time; they may be used when a very short survival is contemplated. Massive homogenous grafts fuse very slowly and always undergo aseptic necrosis with severe deformity. The experimental work of Herndon (1952) and the clinical work of Lexer (1925) showed clearly that they cannot give a lasting functional result.

The only safe procedures are those that bridge the gap caused by the resection with an autogenous graft strong enough to allow weight bearing.

Bridging by a simple or double tibial graft has been used by many authors. The cases published by Phemister (1945), Delannoy, Vernaeghe and Lagache (1950), Delannoy and Soots (1957) and Decoulx and Decoulx (1950) showed a great number of mishaps: non-union, fracture of the graft, with many iterative operations, very prolonged after-care, and a high proportion of failure. Juvara (1921) advised a massive graft obtained by frontal hemisection of the femur (for tumour of the tibia), or of the tibia (for tumour of the femur), but, even with this massive graft, failures are frequent. Twenty-four cases have been published, among which the late results are given only for nine. In one case (Grigorescu and Vasiliu 1934) union was present after five months; in seven cases the delays in obtaining bony union varied between two and five years, with a great number of operative attempts (Juvara 1921, Guilleminet 1950, Montant 1928, Operti 1954). These difficulties and failures probably explain why so few cases of resection are published.

FIG. 1

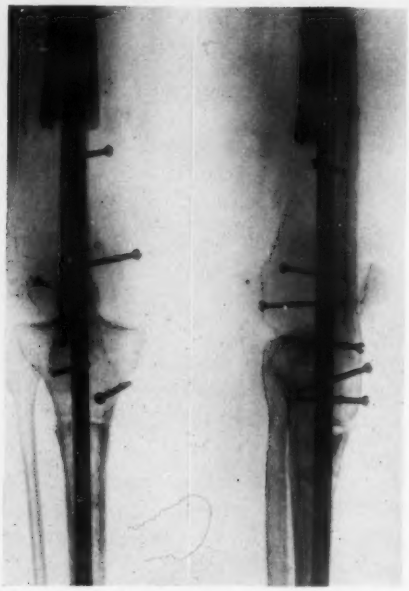

FIG. 2

Case 3—Recurrent giant-cell tumour of lower end of femur. Figure 1—Before excision. Figure 2—Four months after operation (excision, nail, tibial graft, acrylic prosthesis).

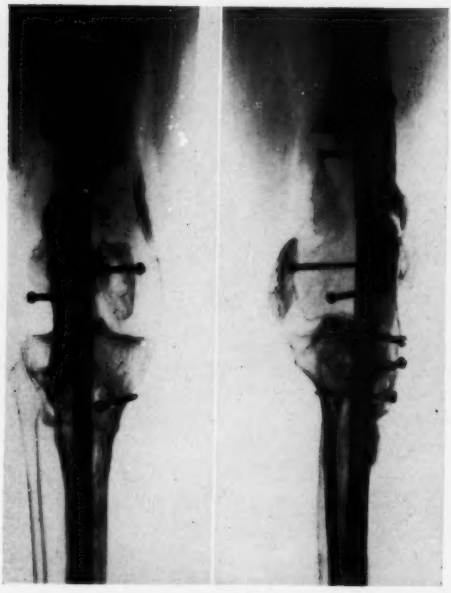

FIG. 3

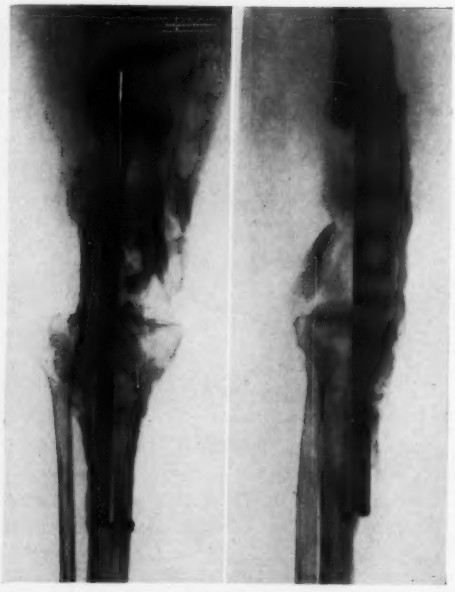

FIG. 4

Case 3. Figure 3—Three years after operation: fracture of graft and nail. Figure 4—Solid union eight months after heterogenous graft.

The procedure described in this paper aims at preventing non-union and fracture of the graft by adding to the Juvara massive graft an intramedullary nail and a massive homogenous graft filling the defect. Furthermore, in order to provide a quicker incorporation of the graft, the back of the patella is freshened and fixed to it; the preserved muscular pedicle of the patella provides a supplementary blood supply to the middle of the graft. This complete procedure was used only in the last five cases.

CASE REPORTS

The records of the patients show the progressive evolution of the technique.

ROTATION TIBIAL GRAFT AND NAIL

Case 1. *Osteolytic sarcoma of the lower end of the femur*—Woman aged forty-five years, accountant. December 1950—Resection of the lower third of the femur together with the patella: intramedullary nailing, bridging by the half tibia. Bony union after eight months. Walked with crutches. October 1952—Pulmonary metastasis; death. *Necropsy*—Local recurrence, multiple metastases, but solid union of the graft.

ROTATION TIBIAL GRAFT, INTRAMEDULLARY NAIL, ACRYLIC PROSTHESIS BETWEEN STUMP OF FEMUR AND UPPER END OF TIBIA

Case 2. *Chondrosarcoma with fracture of the lower end of the femur*—Man aged forty-one years, trader, in bad general state. March 1951—Wide resection, intramedullary nail, rotation tibial graft and acrylic prosthesis. Death the same day from cardiac failure.

Case 3. *Osteolytic giant-cell tumour of the lower end of the femur* (Figs. 1 to 4)—Married woman aged thirty-two. February 1949—Curettage. July 1949—Recurrence; radiotherapy (4,000 r). Patient seen in May 1951—Nearly complete destruction of the lower end of the femur with a pathological fracture. Wide resection of the lower end of the femur: rotation tibial graft, intramedullary nailing, acrylic prosthesis. September 1951—Bony union. Walked with weight bearing for three years. October 1954—Fracture of the nail and the graft; further operation, heterogenous graft; infection. March 1955—Removal of the heterograft, but bony union of the first graft. May 1958—Patient free from recurrence and walking freely.

In this case the fracture of the graft occurred exactly at its middle, at the level of the patella.

ROTATION TIBIAL GRAFT, INTRAMEDULLARY NAIL, ACRYLIC PROSTHESIS, PATELLA FIXED ON THE GRAFT TO ACCELERATE REVASCULARISATION AND PREVENT THE FRACTURE

Case 4. *Osteolytic giant-cell tumour of upper end of tibia*—Man aged twenty-five, shopkeeper. June 1954—Local excision with reconstruction by autograft (giant-cell tumour, grade II of Lichtenstein). September 1954—Local recurrence. October 1954—Wide resection of the upper end of the tibia. April 1955—Local recurrence: amputation through the thigh. March 1957—Death with pulmonary metastases.

ROTATION GRAFT, INTRAMEDULLARY NAIL, HOMOGRAFT IN THE DEFECT, PATELLA FIXED TO AUTOGRAFT

Case 5. *Giant-cell tumour of upper end of tibia* (Figs. 5 and 6)—Man aged forty-two, clerk. 1949—Curettage. 1951—Recurrence: no treatment. 1954—Radiotherapy. Patient seen in March 1955—Recurrence of the tumour: resection of the upper end of the tibia, rotation femoral graft, intramedullary nailing, homoplastic graft in the defect, fixation of the patella. June 1955—Bony union: walks with weight bearing. April 1958—Patient symptomless.

Case 6—*Fibrosarcoma of lower end of femur*—Schoolgirl aged sixteen years. First symptoms in 1953. Patient seen in October 1955: large tumour with spontaneous fracture. Resection of the lower third of the femur: tibial graft, intramedullary nailing, homograft, fixation of the patella. Bony union and walking with weight bearing after four months. April 1957—Local and limited recurrence removed. September 1957—Well. March 1958—Amputation.

FIG. 5

FIG. 6

Case 5—Recurrent giant-cell tumour of tibia. Curettage and x-ray therapy five years previously. Figure 5—Before excision. Figure 6—Good result two and a half years after operation (excision, nail, tibial graft, homogenous graft, fixation of patella).

FIG. 7

FIG. 8

Case 8—Rapidly growing giant-cell tumour of lower end of femur. Figure 7—Before operation. Figure 8—Eight months after operation (excision, nail, autogenous tibial graft, homogenous graft, fixation of patella).

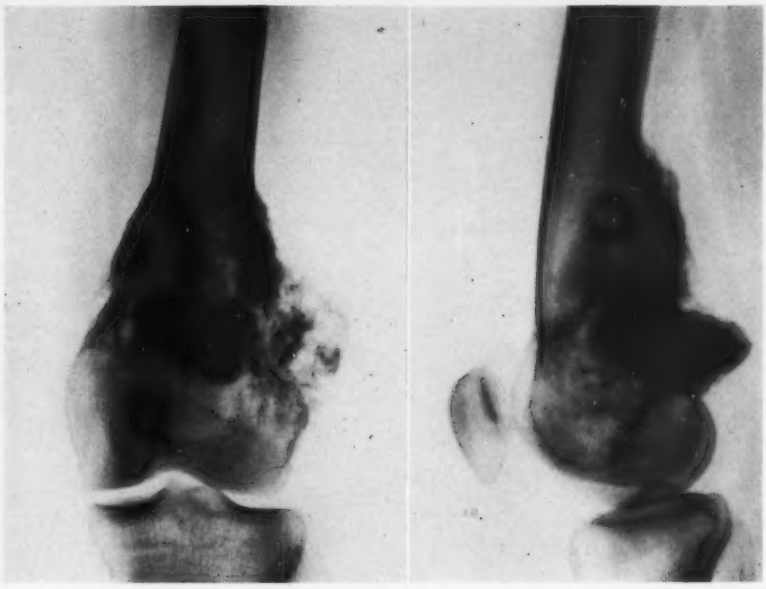

FIG. 9
Case 9—Periosteal sarcoma of lower end of femur. Radiograph before operation.

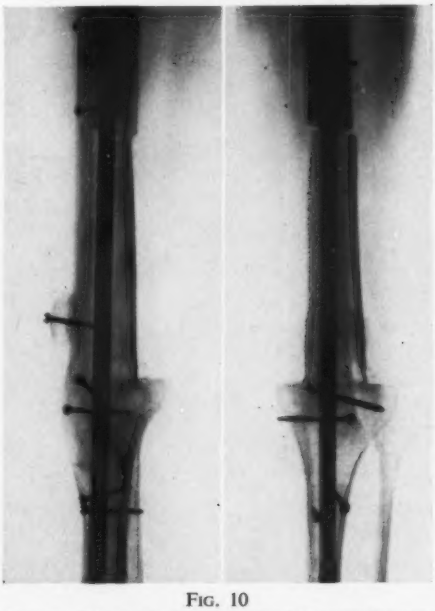

FIG. 10

FIG. 11

Case 9. Figure 10—One month after operation (excision, nail, autogenous graft, homogenous graft, fixation of patella). Union occurred but graft broke later. Figure 11—After regrafting.

Case 7. *Osteolytic infected giant-cell tumour of lower end of femur*—Married woman aged twenty-five years. February 1956—Curettage and radiotherapy. Patient seen in July 1956—Bone cavity infected; biopsy showed recurrence of the tumour. September 1956—Resection of the lower end of the femur. Immobilisation in plaster with two Steinmann pins; suction drainage of the cavity and antibiotics. October 1956—Wound closed; no discharge. Bridging of the gap by tibial graft, intramedullary nail and homograft, fixation of the patella. Bony union in three months, but recurrence of infection. Nail removed together with homograft. Fracture of the autograft. Amputation.

Case 8. *Osteolytic fast-growing giant-cell tumour of lower end of femur, clinically malignant* (Figs. 7 and 8)—Girl aged seventeen, dressmaker. Biopsy: giant-cell tumour, grade II of Lichtenstein. February 1957—Radiotherapy. March 1957—Resection of the lower end of the femur: rotation tibial graft, intramedullary nailing, homoplastic graft, fixation of the patella. Bony union; walks with weight bearing after three months. June 1958—Well.

Case 9. *Periosteal sarcoma of lower end of femur* (Figs. 9 to 11)—Married woman aged twenty-eight years. November 1956—Resection of the lower end of the femur: rotation tibial graft, intramedullary nailing, homoplastic graft, fixation of the patella. Walked with weight bearing after four months. June 1957—Fracture at the upper part; new graft. May 1958—Walking freely.

RESULTS

This series allows a conclusion only on the value of the operative technique used for re-establishing continuity of the bone (Table I).

TABLE I

CLINICAL DETAILS AND RESULTS (NINE CASES)

Case number	Age (*years*)	Diagnosis	Type of operation	Early result	Late result
1	45	Osteosarcoma of femur	Excision. Tibial autograft and nail	Bony union	Death from dissemination at two years
2	41	Osteosarcoma of femur	Excision. Tibial autograft, nail, acrylic prosthesis	Death first day	—
3	32	Giant-cell tumour of femur	Excision. Tibial autograft, nail, acrylic prosthesis	Fracture of graft and nail after three years. Heterograft	United and symptom-free at seven years
4	25	Giant-cell tumour (grade II) of tibia	Excision. Femoral autograft, nail, acrylic prosthesis	Recurrence: amputation	Pulmonary metastases at three years; died
5	42	Recurrent giant-cell tumour of tibia	Excision. Femoral autograft nail, massive homograft, and patella	Union. Walked at three months	Symptom-free at three and a half years
6	16	Fibrosarcoma of femur	Excision. Tibial autograft, nail, massive homograft and patella	Union. Walked at four months	Recurrence after two years: amputation
7	25	Infected recurrent giant-cell tumour of femur	Excision. Tibial autograft, nail, massive homograft and patella	Union but infection recurred	Fracture of graft at eight months. Amputation at one year
8	17	Giant-cell tumour (grade II) of femur	Excision. Tibial autograft, nail, massive homograft and patella	Union	United and symptom-free at fifteen months
9	28	Periosteal sarcoma of femur	Excision. Tibial autograft, nail, massive homograft and patella	Union. Walked at three months	Fracture of graft at eight months. United and symptom-free at one and a half years

Early results (Table II)—In this series of nine patients one died on the day of the operation (Case 2), one had an early recurrence and underwent amputation (Case 4), one with infection also required amputation. All the others showed union and were able to walk freely with weight bearing after three or four months, but two of them sustained a secondary fracture

TABLE II

EARLY RESULTS (NINE CASES)

Technique	Number of cases	Result
Nail, half tibia or femur, and acrylic prosthesis	4	1 death. 3 bony union (but 1 fracture of graft and nail after 3 years)
Nail, half tibia or femur, massive homogenous graft, and patella fixed to auto-graft	5	1 recurrence of infection with fracture of graft—amputation. 4 bony union with weight bearing before 3 months (1 fracture of graft at 7 months regrafted and united)

of the graft and underwent regrafting, and one patient had a recurrence and has had the limb amputated.

Four patients now show strong bony continuity between the femur and the tibia, after seven years, three and a half years, eighteen months and fifteen months.

Late results (Table III)—Late results must be considered separately for sarcoma and giant-cell tumour.

Sarcoma—The results are bad. Out of four cases we find: one operative death (Case 2), one generalisation after two years (Case 1), one local recurrence after two years (Case 6), one patient symptomless after eighteen months (Case 9).

Giant-cell tumour—All the cases of this series were severe: three patients were treated for recurrence after curettage and radiotherapy (Cases 3, 5, 7); one patient's tumour was growing

TABLE III

LATE RESULTS (NINE CASES)

Diagnosis	Number of cases	Result
Sarcoma	4	1 early death. 1 generalisation at 8 months. 1 recurrence at 2 years; amputation. 1 symptom-free at 1 year
Giant-cell tumour (all recurrent and extensive)	5	1 recurrence; amputation; pulmonary metastasis. 4 free from recurrence after 7 years, 3½ years, 1 year, 15 months

rapidly and was clinically, radiologically and histologically suspect of malignancy (Case 8). These four patients are free from recurrence after seven years (Case 3), three and a half years (Case 5), one year (Case 7) and fifteen months (Case 8). One patient (Case 4) showed a malignant evolution of the tumour, with recurrence after local excision and after resection, and died from metastases after amputation.

OPERATIVE TECHNIQUE

Resection for tumour of the lower end of the femur (Fig. 12)—The patient is supine. A tourniquet is used. The skin incision is postero-lateral on the lower part of the thigh, antero-lateral on the knee and anterior on the leg. The tumour is dissected on its lateral and anterior aspects. The knee joint is widely opened. The ligaments, including the patellar ligament, are cut. The femoral shaft is divided four or five centimetres above the upper limit of the tumour. Dissection of the femur is completed; it is made easier by mobilisation of its lower end. Particular attention is given to the femoral vessels, but the dissection must be made at a distance from the tumour in sound muscular tissue. The lower end of the femoral shaft is cut in steps five or six centimetres long. The tibia is divided with the electric saw into two

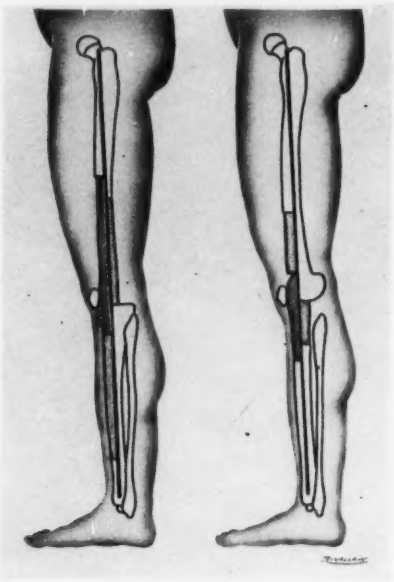

FIG. 12

FIG. 13

Figure 12—Diagrams showing the operation. *Left*—Bridging of femoral defect by a tibial graft and bone bank graft, with fixation of patella. *Right*—Bridging of tibial defect by a femoral graft. Figure 13—Line of skin incision.

halves in a frontal plane, for its whole length except the lowest six or seven centimetres. A homogenous tibial or femoral graft is prepared; its length must be exactly the same as the defect between the femur and the tibia. Its anterior aspect is freshened. A long Küntscher nail must be available—seventy to eighty centimetres long according to the length of both tibia and femur, and nine to eleven millimetres in section according to the width of the medullary canals. This nail is hammered into the femoral medullary canal from the lower end upwards, brought out through the buttock, and then hammered down through the medullary canal of the homograft and the tibia. The massive tibial graft is fixed upside down by screws to the lower end of the femoral shaft, to the graft and to the upper end of the tibia. A homogenous graft may be fixed to reinforce the upper part of the tibia. The back of the patella, which has been preserved with its muscular attachment, is freshened and screwed to the front of the tibial autograft as close as possible to the middle of the defect. The skin is closed over a suction drain.

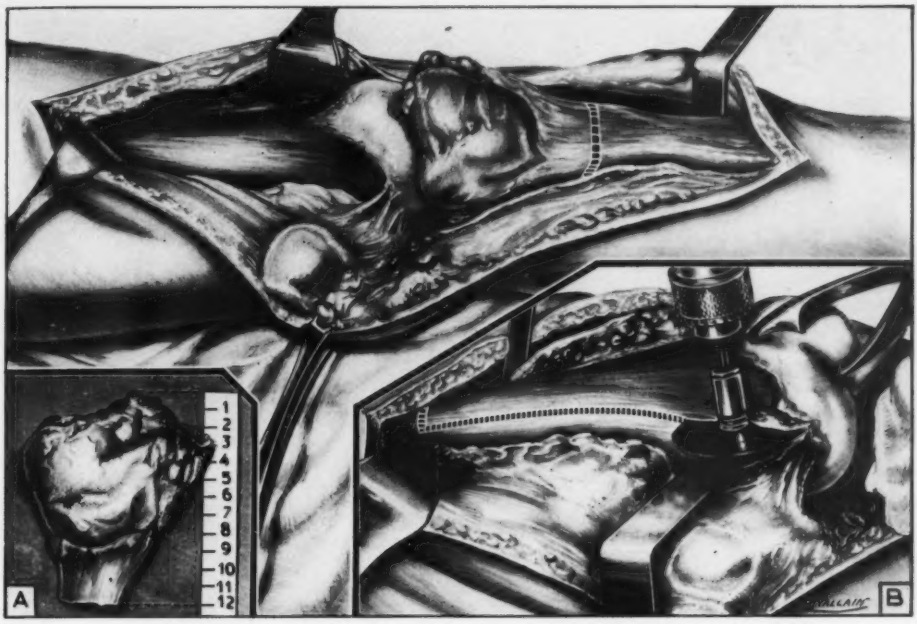

FIG. 14

Top—Approach to the tumour and lower end of femur. The patella is dissected with its muscular pedicle.
Bottom—Taking the femoral graft.

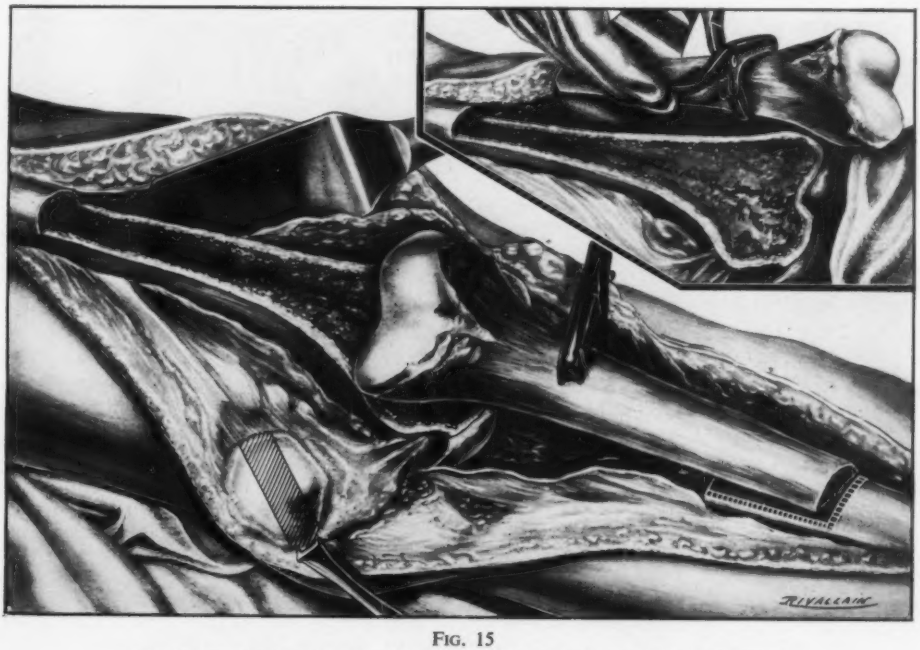

FIG. 15

The femoral graft is turned upside down. The back of the patella is freshened.

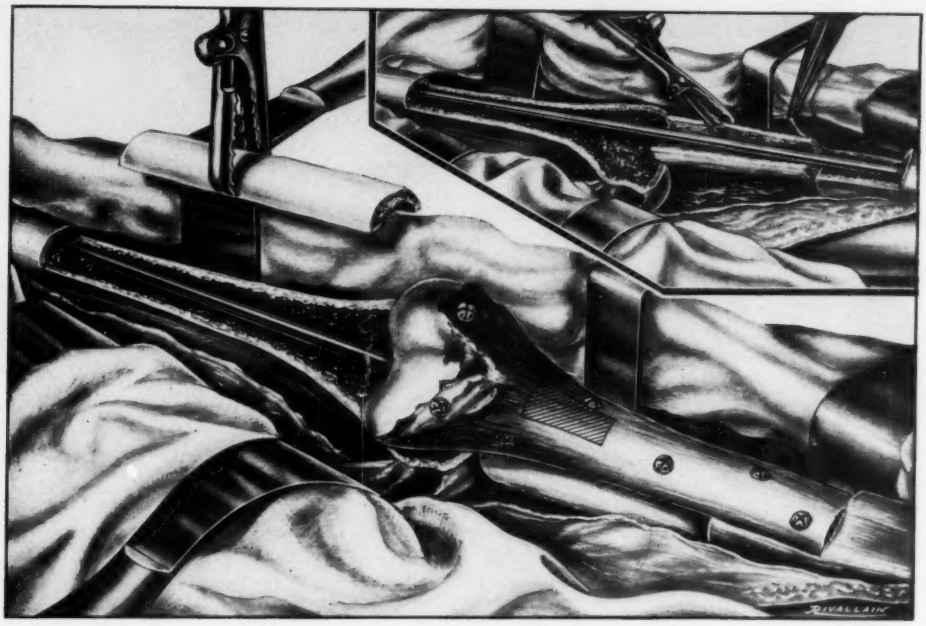

FIG. 16

Top—A homograft is inserted in the defect. *Bottom*—Intramedullary nailing. The femoral graft is fixed by screws to the femur, tibia and homograft. The femur is reinforced by a homograft. The femoral graft is freshened for fixation of the patella.

Resection of the upper end of the tibia (Figs. 13 to 16)—The operative technique is the same but reversed. Dissection of the tumour is accomplished after division of the shaft and opening of the joint. The massive graft is obtained by frontal hemisection of the lower part of the femur.

CONCLUSION AND SUMMARY

1. An operation to re-establish the continuity of the bone after resection of tumours of the lower end of the femur or upper end of the tibia is described.
2. Sound bony union may be obtained, allowing walking with weight bearing after three to four months.
3. The combination of a massive tibial or femoral graft (Juvara procedure) with an intramedullary nail, a massive homogenous graft, and fixation of the patella to the autograft to accelerate its revascularisation, has given constant and rapid bony union in five cases.
4. This possibility gives real value to wide resection in the treatment of severe cases of giant-cell tumours, especially those that are recurrent, osteolytic or suspect of malignancy.
5. Its value in the treatment of sarcoma cannot be assessed in so short a series.

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NON-UNION OF THE EPIPHYSIS OF THE LATERAL CONDYLE OF THE HUMERUS

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It is well known that a fracture-separation of the epiphysis of the lateral condyle of the humerus occasionally fails to unite. Radiographs showing non-union long after the causative injury are featured prominently in most text-books of orthopaedics. The patients are usually seen at our clinics either because of the onset of a tardy ulnar paresis, or because a youth's parents are anxious to know whether their son's mobile but somewhat unsightly valgus elbow is likely to render him unfit for a permanent commission in one of Her Majesty's Services.

It is not in doubt that these lesions result from the failure of a displaced main lower humeral epiphysis to unite or that the displacement results from injury. Indeed, there is almost always a history of an injury to the elbow between the ages of four and ten, though there is seldom anything to suggest that the injury had been of more than minor severity, in spite of the markedly abnormal radiograph frequently present at the time of attendance. Evidence will be presented which indicates that most of the cases of non-union seen in this country arise as the late results of quite minor epiphysial displacements which have progressed unrecognised to non-union, often with increasing deformity, rather than from untreated major displacements. It has long been widely recognised that the characteristically grossly displaced and rotated epiphysis (Fig. 4) will almost inevitably fail to unite with the diaphysis if not dealt with by open reduction. For this reason non-union from the neglect of such cases is uncommon where orthopaedic and accident services are well developed and within easy reach of the general population.

It is necessary briefly to review the pathological anatomy of the epiphysial displacement, detailed descriptions of which have been published by Speed and Macey (1933), Wilson (1936), Kini (1942), Smith and Joyce (1954), and Watson-Jones (1955). The line of separation of the fragment is at the epiphysial line, except postero-laterally where a wedge of diaphysial bone of variable size separates with the epiphysis. The separated fragment represents about half of the lower articular surface of the humerus.

Displacement can most conveniently be divided into two degrees: 1) *minor*, when the epiphysis is displaced slightly outwards and perhaps also tilted a little (Fig. 1); and 2) *major*, when the epiphysis is completely dislocated. Here there may be gross rotational displacement in addition to wide lateral shift—described by some writers as somersaulting (Fig. 4)—or there may be backward displacement of the epiphysis together with the forearm bones (Fig. 6). The injury is usually seen between the fourth and tenth years, though I have observed it in children as young as two and as old as fifteen.

Failure of union has been observed in three differing sets of circumstances, as follows. **Non-union after operative reduction**—Union did not occur in three cases in which major displacement had been reduced by open operation and secured by soft-tissue suture. This is an accepted method of treatment which is usually considered to give satisfactory results (Watson-Jones 1955). Nevertheless, reductions held by soft-tissue suture are usually not exact, nor so firmly held, as when internal fixation by a screw is used. In none of the three cases of non-union in this group was the surgeon who had treated the case aware that there had been a failure of union. Radiographs soon after operation had been deemed satisfactory, and restoration of movement had made good progress at an early phase. Figures 10, 11 and 12 illustrate one of these cases. The grossly displaced and rotated epiphysis (like that in Figure 4) was dealt with by open reduction and soft-tissue suture. Reduction was imperfect, as shown in Figure 10. Radiographs taken after one month (Fig. 11) showed a deterioration in position.

FIG. 1

Case 1—Minor displacement, consisting in lateral shift and slight tilting but no rotation of the fragment. The diaphysial fragment which separates with the epiphysis is well shown.

FIG. 2

FIG. 3

Case 1. Figure 2—Five months after injury. No union. Treatment had been by closed reduction and five weeks in a long arm plaster. Figure 3—Six months after bone grafting by method described. Sound union present.

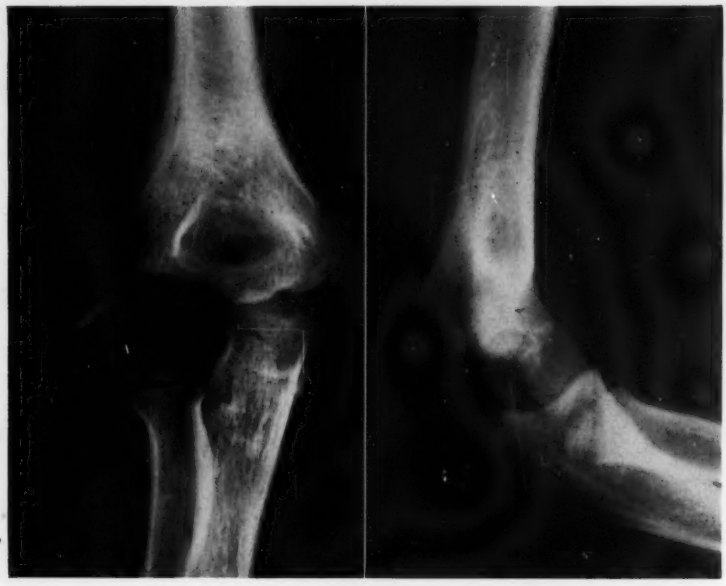

FIG. 4

Case 2—Major displacement: complete dislocation of the epiphysis with marked rotational displacement of the lateral condyle.

FIG. 5

Case 3—A fracture-separation like that shown in Figure 4, after open reduction and screw fixation. The screw is passed through the fragment of diaphysial bone which separates with the epiphysis, at a right angle to the fracture line. The screw avoids the epiphysis.

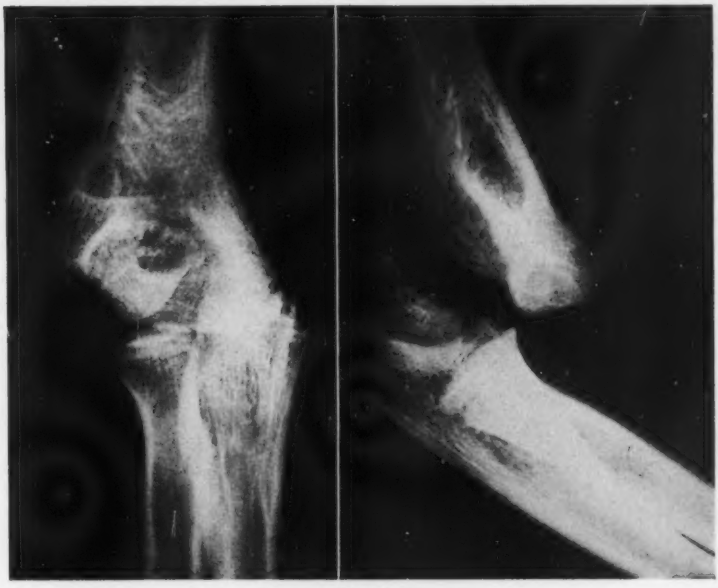

FIG. 6

Case 4—Major displacement with complete backward dislocation of the epiphysis. The forearm bones dislocate backwards with the lateral condyle.

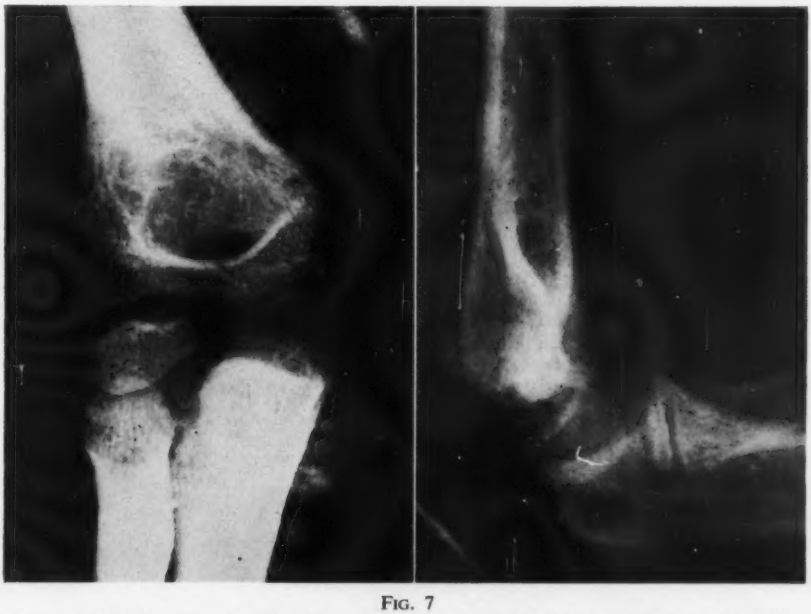

FIG. 7

Case 5—A fracture-separation like that shown in Figure 6, after closed reduction. This position was accepted.

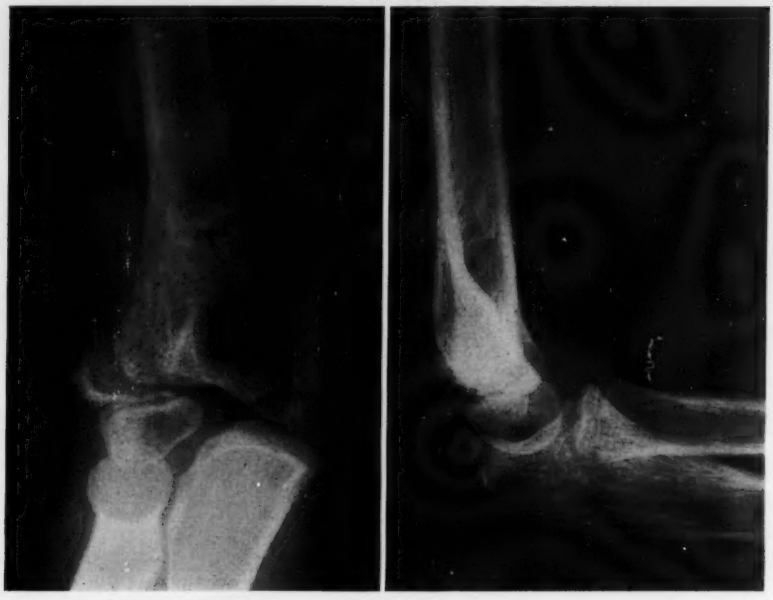

FIG. 8
Case 5—No evidence of union three months after fracture. Six weeks' immobilisation in a long arm plaster was used after closed reduction.

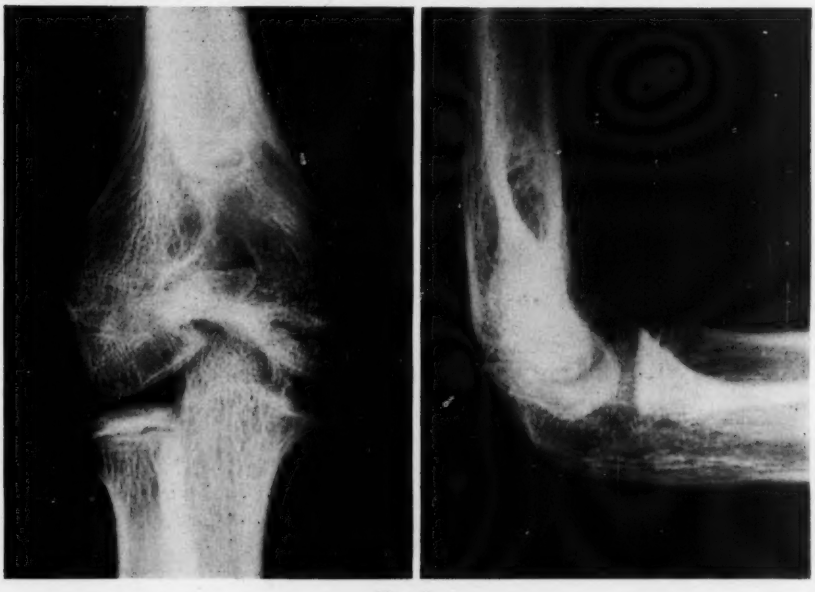

FIG. 9
Case 5—Three years after bone grafting by the technique described.

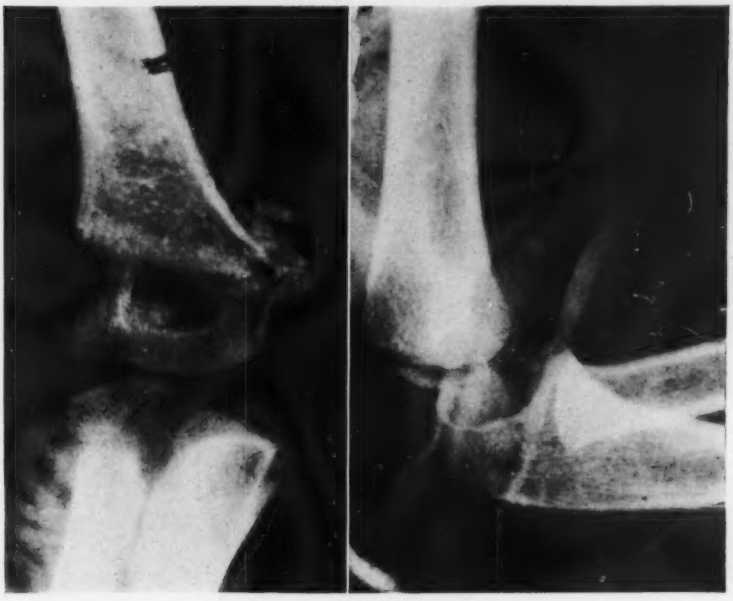

FIG. 10

Case 6—A case of major displacement (like that shown in Figure 4) immediately after open reduction and soft-tissue suture. Reduction is imperfect, as is often the case after soft-tissue suture.

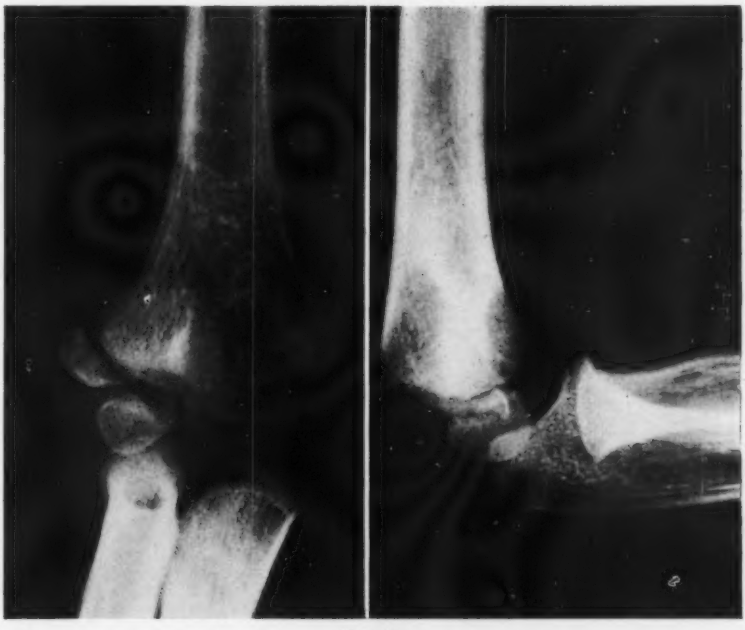

FIG. 11

Case 6—One month after open reduction. Increased lateral luxation of the epiphysis is shown.

FIG. 12

Case 6—Eight years later, showing non-union. There was a valgus deformity of the elbow.

FIG. 13

FIG. 14

Case 7. Figure 13—Epiphysial separation without displacement. Figure 14—One year later: non-union. Treatment had been by support with a sling for three weeks. Movements rapidly returned to normal.

FIG. 15

Antero-posterior radiographs of the elbows of six patients with lateral condylar epiphysial non-union who were treated by ulnar transposition for tardy ulnar palsy. In the starred case (*bottom left*) non-union followed an open reduction and soft-tissue suture. The other five are considered to have followed minor displacements. In all six cases the relationship of the capitulum with the radial head is such as could not have resulted if the lateral condyle had been completely dislocated and left unreduced. (It will be seen that ulnar palsy can be associated with only slight deformity of the elbow. Onset of symptoms of ulnar palsy in this group varied from three to thirty-nine years after the injury.)

Immobilisation was then discontinued. The patient was discharged six months later with almost full movement and no symptoms. Further radiographs were not taken. After another eight years he reported with full movement but a valgus elbow and unequivocal non-union (Fig. 12).

Non-union after closed reduction—Non-union has twice occurred in patients treated by closed reduction and plaster splintage under my own supervision. The first, a case of relatively minor displacement (Fig. 1), was dealt with by manipulation and the application of a moulded long arm plaster. Immobilisation was continued for five weeks. Radiographs showed that

union was not established at five months (Fig. 2). Presumably the impossibility of holding the displaced fragment of bone firmly and exactly in position by plaster, after reduction by direct digital pressure, is a major factor in determining the non-union in such cases, though we have all seen similar examples where union in good position has become established. Wilson (1955) illustrated an almost identical case. His patient developed increasing displacement and deformity, and the radiographs four years after injury showed non-union and a grossly valgus elbow. My second case was one in which a major posterior dislocation of the epiphysis accompanied by backward displacement of the forearm bones (like that in Figure 6) was reduced by manipulation. The reduction was imperfect but was regarded as satisfactory (Fig. 7). Union did not become established in spite of six weeks' immobilisation in a long arm plaster (Fig. 8).

Non-union after undisplaced fracture-separations—Non-union became established in such a case while the patient was under my observation. The original radiograph showed a mere fissure of the bone (Fig. 13). Treatment was by support with a sling for three weeks. Symptoms soon disappeared and movements rapidly returned to normal, but later radiographs showed failure to unite and lateral displacement of the epiphysis (Fig. 14).

Study of the radiographs of six patients with non-union of the lateral condyle who were treated for ulnar neuritis by anterior transplantation of the ulnar nerve at the Princess Elizabeth Orthopaedic Hospital, Exeter (Fig. 15), showed that five almost certainly resulted from minor displacements. The sixth was the sequel of an open reduction and soft-tissue suture in which union had not ensued. In all six cases the relationship of the capitulum with the radial head was such as could not have resulted if the lateral condyle had been completely dislocated and left unreduced. This group of transplantations represented 10 per cent of the ulnar transpositions carried out at Exeter between 1947 and 1956.

PREVENTION AND TREATMENT OF NON-UNION

Following the above findings certain recommendations are made.

First, when open reduction is carried out the displaced fragment should be stabilised if possible by internal fixation with a screw. The screw should be inserted so as to avoid the epiphysis, as illustrated in Figure 5. It is contended that screw fixation provides an accurate and stable reduction which cannot be matched by soft-tissue suture. This technique should not only reduce the risks of non-union but also eliminate the well known phenomenon of "fishtailing" so often seen after soft-tissue suture. The highest possible standard of reduction should be regarded as mandatory in a lesion that divides an articular surface of a major bone of a child into roughly equal parts. It is not denied that the insertion of the screw is awkward, but it is considered that it is a well worth while procedure. In very young children the ordinary bone screws cannot be used because they are too thick: in such cases a bone nail or Kirschner wire provides a satisfactory substitute.

Secondly, imperfect closed reductions must be critically assessed with a strong bias towards open reduction and screwing. Some authorities have recommended open reduction and internal fixation in all cases in which significant displacement has occurred (Speed and Macey 1933, Wilson 1936). I agree with those who are willing to attempt closed reduction if there is no rotation of the epiphysis, in order to avoid, if possible, an operation scar which grows with the child (Watson-Jones 1955, Wilson 1955, Smith and Joyce 1954).

Thirdly, union of displaced lower humeral epiphyses must be established unequivocally by radiographs. If adequate radiographic studies are insisted upon delayed union can be recognised and steps can be taken to expedite union, so preventing the increasing displacement and deformity that may otherwise occur. In particular, frequent check radiographs are necessary after closed reductions to exclude redisplacement during the period of immobilisation.

Fourthly, when delayed union is present and when, in children, non-union without major displacement has become established, it is suggested that bone grafting be carried out by

drilling through the fragment of the diaphysis which separates with the epiphysis and on into the main part of the diaphysis. The drill track is then packed with iliac cancellous bone. Plaster immobilisation is continued until union has occurred. This method was used successfully in the three cases of non-union which arose under my supervision (Figs. 3 and 9).

In all, thirteen cases of non-union of the humeral lateral condylar epiphysis have been studied. Three followed open reduction and soft-tissue suture. The remaining ten followed minor displacements. There was no example of non-union after a major displacement which had remained untreated. Such cases would inevitably result in non-union but they are unlikely to be seen where efficient orthopaedic and accident services are within easy reach of the general public.

SUMMARY

1. Thirteen cases of non-union of the epiphysis of the lateral condyle of the humerus were studied. Ten followed minor lateral luxations of the epiphysis; three were sequelae of open reduction and soft-tissue suture of major displacements.

2. In three cases in which non-union developed while the patient was under observation, union was secured after bone grafting by a technique described.

I wish to thank my colleagues at the Princess Elizabeth Orthopaedic Hospital for allowing me to study their cases. My thanks are also due to Mr Arthur Reaney for the radiographic reproductions.

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TUBERCULOSIS OF THE FEMORAL NECK AND GREATER TROCHANTER

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TUBERCULOSIS OF THE FEMORAL NECK

During the nine years up to the end of 1956 six cases of tuberculosis of the femoral neck have been treated at Heatherwood Orthopaedic Hospital. This relatively uncommon lesion has formed less than 0·5 per cent of all the skeletal tuberculosis treated.

Many authors have regarded tuberculosis of the femoral neck as the primary site of bone involvement in tuberculosis of the hip joint, and have included it in reviews of that lesion. This is not surprising because, in the absence of chemotherapy, most of these lesions have extended into the hip joint to cause a pan-arthritis irrespective of the method of treatment.

Bergman (1930) described a good result in a single case with a juxta-epiphysial lesion treated conservatively. He also quoted four cases of Waldenstrom's with satisfactory results two years after treatment by excision. Logroscino (1936) gave a useful classification of lesions of the femoral neck. He divided these on a regional basis into four types of focus: 1) cervico-diaphysial; 2) juxta-epiphysial; 3) bulbo-metaphysial; 4) metaphysial subtrochanteric. He claimed good results in six of fifteen patients treated conservatively and stated that a pan-arthritis followed excision.

Hatcher and Phemister (1937), in a review of tuberculosis of the hip joint in seventy children, stated that the juxta-epiphysial region of the femoral neck was the commonest site of primary bone involvement and was affected in fourteen children. The hip joint remained intact in only two of these patients. Cholmeley (1939) reported fifty-five patients with extra-articular lesions. The disease spread to involve the hip in eleven out of twelve patients treated surgically and forty out of forty-three treated conservatively. Ponseti (1948) referred to seven out of thirty-one patients with para-articular foci. Four were in the ilium and three in the femoral neck. The disease spread to involve the hip in all seven patients irrespective of the method of treatment.

Dobson (1951), in a review of 320 cases of tuberculosis of the hip, stated that primary bone involvement of the femoral neck occurred in only fifteen patients. Wilkinson (1953) stated that a tuberculous infection of the ends of long bones was usually accompanied by involvement of the synovial membrane. He claimed satisfactory results in nineteen of twenty-three patients treated by curettage and partial synovectomy under antibiotic cover.

CLINICAL REVIEW

Three of the six patients under review were treated conservatively. Two of these patients made a complete recovery, but radiologically it was shown that healing was very slow. The third patient treated conservatively developed tuberculous arthritis with destruction of the joint two and three-quarter years later. The remaining three patients were treated by excision of the focus and chemotherapy. These three patients received streptomycin combined with para-amino-salicylic acid (PAS) or izo-nicotinic acid hydrazide (INH) or both. All three made a rapid and complete recovery. The improvement noted radiologically at the end of three months was most convincing.

CASE REPORTS

Case 1—Girl aged three years. A slight limp and discomfort in the region of the right hip joint had been noticed for fourteen months before her admission. There was slight restriction of movement. Radiographs revealed a lesion of the metaphysial subtrochanteric region with involvement of the

FIG. 1

FIG. 2 FIG. 3

Case 3. Figure 1—Radiograph showing large juxta-epiphysial lesion of femoral neck. Figure 2—Two years later: the lesion appears healed. Figure 3—Two and a half years later still: the hip joint is involved.

inner part of the trochanteric epiphysis. She was treated by bed rest and immobilisation in plaster for twelve months. Radiographs then showed recalcification and partial obliteration of the focus. She regained full movement. The focus could still be seen in radiographs taken three years after discharge. When she was last seen nearly seven years after treatment normal function was present at the hip.

Case 2—Girl aged five years. This patient had been seen and investigated elsewhere for discomfort and slight limp at the left hip, and had already undergone eight months' immobilisation in plaster for a lesion of the left femoral neck. There was slight restriction of movement at the left hip, and radiographs revealed an extensive lesion of the cervico-diaphysial region. She was treated by bed rest and immobilisation in plaster for a further fourteen months. Radiographs then showed that the lesion was well localised, with good surrounding calcification. She was allowed up in a weight-relieving caliper which was worn for a further ten months. Full movement was rapidly regained. Radiographs taken six years after discharge showed complete obliteration of the focus. When she was last seen six and a half years after treatment normal function was present at the hip.

Case 3—Boy aged eight years. Discomfort and slight limp at the right hip joint had been noticed for seven months, and movements at the hip joint were restricted to half the normal range. Radiographs revealed an extensive juxta-epiphysial lesion (Figs. 1 to 3). The boy was treated by bed rest and immobilisation in plaster for fifteen months. Radiographs then showed apparent healing with obliteration of the cavity but, clinically, movement was still only through half the normal range. Activity was encouraged, but movement did not improve. Radiographs taken nine months later showed further improvement. Two and three-quarter years later there was increasing pain and stiffness, with flexion-adduction deformity, and radiographs then revealed involvement of the hip joint.

Case 4—Boy aged three years. Discomfort and slight limp at the left hip joint had been noticed for three months, and there was slight restriction of movement. Radiographs revealed an extensive lesion of the femoral neck extending from the trochanteric region across the epiphysial line to involve the inner and lower part of the capital epiphysis (Fig. 4). The diagnosis was supported histologically by an external iliac gland biopsy. No improvement was noted either clinically or radiologically after two months' immobilisation in plaster and chemotherapy. Curettage of the focus through a subtrochanteric approach was performed.

Operation—A large sequestrum was removed. The lesion was very vascular and no pus was encountered. The cavity communicated with the hip joint. No bone chips were used. The cavity was dusted with streptomycin, and the wound closed.

Progress—There was immediate and striking improvement in the patient's general condition (Fig. 5). Immobilisation was continued for three months, and chemotherapy for a total of three and a half months, including the period before operation. Radiographs showed excellent recalcification and obliteration of the cavity (Figs. 6 and 7). Full movement was present at the hip and the patient rapidly regained his activity. The total period of observation was four and a half years, and normal function was present at the left hip joint.

Case 5—Boy aged nine years (Fig. 3). This child developed miliary tuberculosis and proved meningitis at the age of two and three-quarter years, and made a complete recovery after six months' intrathecal and intramuscular streptomycin. Two and three-quarter years later he developed tuberculous arthritis of the left hip and again made a complete recovery with conservative treatment for a year, with a four months' course of streptomycin. He was readmitted over two years later for investigation of discomfort and slight restriction of movement at the right hip which had been present for a month. Radiographs revealed an early lesion of the outer part of the femoral neck (Fig. 8). He was treated by immobilisation and chemotherapy, but radiographs taken five weeks later showed rapid destruction of the femoral neck (Fig. 9). Curettage of the focus through a Smith-Petersen approach was performed.

Operation—The lesion was still separated from the joint by a thin periosteal layer. A large sequestrum with much debris was removed from a large cavity involving three-quarters of the substance of the femoral neck. Erosion had extended to the cartilage of both the capital and trochanteric epiphysial lines. No bone chips were used. The cavity was dusted with streptomycin powder and the wound closed.

Progress—Immobilisation was continued for three months and chemotherapy for a total of four months including the time before operation. Radiographs then showed marked improvement in density and definition, with filling in of the cavity (Fig. 10). Full movement was present. The patient has been observed for two and a half years, and normal function is present at the hip. Culture and guinea pig inoculation of material from the bone cavity were negative.

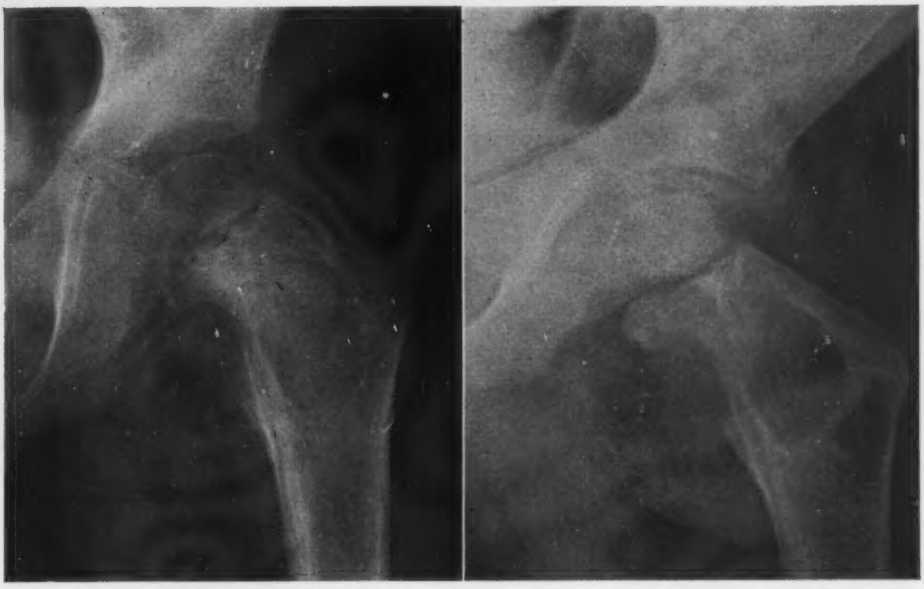

FIG. 4 FIG. 5

Case 4. Figure 4—Radiograph showing large focus in femoral neck with sequestration and involvement of the lower part of the capital epiphysis. Figure 5—Appearance after operation, a month later.

FIG. 6 FIG. 7

Case 4. Figure 6—Three months after operation, showing excellent recalcification and obliteration of cavity. Figure 7—Appearance one year and seven months after operation.

FIG. 8 FIG. 9

Case 5. Figure 8—Radiograph showing an early lesion of the outer portion of the femoral neck. Figure 9—Five weeks later, showing sequestration and rapid spread of lesion.

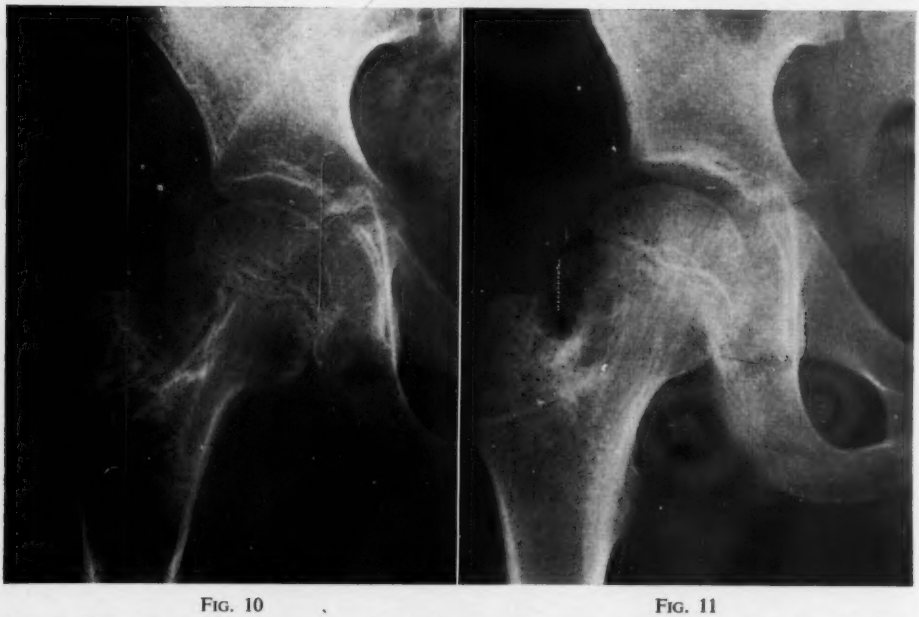

FIG. 10 FIG. 11

Case 5. Figure 10—Condition at four months. Figure 11—Condition at nine months: the lesion is healing.

Case 6—Boy aged three years. He had been investigated nine months before for a left-sided limp; no diagnosis was made. A cold abscess was present a month before his admission. Full movement was present at the left hip. Radiographs revealed a lesion in the distal part of the left femoral neck. He was treated by immediate curettage of the focus through a subtrochanteric approach.
Operation—A large abscess cavity deep to the fascia lata was curetted and was shown to communicate with the focus anteriorly. Sequestra were removed. No bone chips were used. The cavity was dusted with streptomycin and the wound closed.
Progress—Immobilisation in plaster and chemotherapy were continued for three months. Radiographs then showed excellent recalcification and filling in of the cavity. Full movement was present at the hip. Nine months later there was normal function at the hip. Culture of the material from the bone cavity grew tubercle bacilli.

CONCLUSIONS

A study of these six patients suggests that immediate evacuation of the focus combined with chemotherapy is the satisfactory treatment of this lesion. Radiological evidence of healing, even as early as three months after operation, was most convincing, and compared favourably with the much longer process in the two patients who eventually recovered under conservative treatment.

A lesion which radiologically appears to have come to the surface and to communicate with the joint may not do so (Case 5). The prognosis, even when a lesion communicates with the joint may still be good (Case 4). There is probably a critical interval between infection of a joint cavity from this type of lesion and the stage when hyaline cartilage is irretrievably damaged. Surgery and chemotherapy during this interval can still preserve normal function. Cavities situated in the distal part of the neck can be emptied successfully through a subtrochanteric approach without opening the joint.

In all of the three patients treated by chemotherapy and operation the lesion appeared active and the cavity showed no surrounding sclerosis. The vascularity of these lesions was thought to be important in favouring rapid healing. The patient in Case 3, who never had good movement at any stage, probably had a concomitant synovial involvement of the hip joint.

SUMMARY

Tuberculosis of the femoral neck is an uncommon lesion occurring in the first decade. In three cases treated by immediate surgery under antibiotic cover excellent results were obtained.

TUBERCULOSIS OF THE TROCHANTERIC BURSA AND GREATER TROCHANTER

During the seventeen years up to the end of 1956 thirty-two patients suffering from tuberculosis of the gluteal femoral bursa and greater trochanter have been treated in Heatherwood Orthopaedic Hospital. The treatment and results are reviewed.

Chemotherapy has profoundly influenced the treatment and prognosis of this condition. Eighteen of the patients treated during the last seven years of the period under review were subjected to a standardised procedure consisting of radical surgery and chemotherapy. The results are compared with those obtained in the fourteen patients treated earlier, for whom different methods were employed.

There is now an extensive literature on trochanteric tuberculosis. Teale first described the condition as a clinical entity in 1870. Meyerding and Mroz (1933) reported nineteen cases, all proved: two had bursal involvement alone and fourteen had tuberculosis elsewhere. Sinuses persisted in four after radical excision. Wassersug (1940) reported eighteen cases. Two had bursal involvement. Five showed no evidence of tuberculosis elsewhere. Ten of these patients were subjected to radical surgery and in eight the lesion was healed after three years. Alvik (1949) reported thirty-six patients treated between 1936 and 1948. He believed that the disease spread from bursa to bone, and that the bursa might become infected by

TABLE I

CLINICAL DETAILS IN EIGHTEEN CASES OF TUBERCULOSIS OF THE TROCHANTERIC BURSA AND GREATER TROCHANTER TREATED BY THE "STANDARD" METHOD DESCRIBED IN THE TEXT

Case number	Sex	Age (*years*)	Side	History	Other lesions		Result	Confirmatory findings	Comment
					Site	Time relationship			
Infection of bursa alone									
7	F	42	L	Swelling and tenderness 6 months	Lung	Before	Healed 1½ years	Pus TB +ve	Only case with early bursal involvement
8	M	39	R	Small superficial abscess 1 month	Lumbar spine	Before	Healed 3½ years	Histology +ve	—
9	M	21	L	Small superficial abscess 1 month	Left knee	Before	Healed 3¾ years	Histology +ve	—
10	F	67	L	Aching and stiffness 6 months. Superficial abscess 1 month	—	—	Healed 2 years	Histology +ve	—
11	M	24	L	Slight discomfort 7 years. Swelling 3 years: abscess incised: sinus 3 months	—	—	Healed 1¼ years	Pus TB +ve	Grossly thickened and diseased bursa
12	M	20	L	Sinus 8 years before. Healed: reformed 5 months	Lung	Before	Healed 2 years	Pus TB +ve	—
13	M	25	L	Discomfort 2½ years. Abscess 5 months	Lung	Before	Healed 6¼ years	Histology TB +ve Guinea pig +ve	—
14	M	56	R	Sinus 5 years before. Healed: reformed 3 months	Lumbar spine	Before	Healed 3¼ years	Histology +ve	—
Infection of bursa and trochanter									
15	M	37	R	Recurrent sinus formation 18 years: 4 unsuccessful operations	—	—	Healed 2½ years	Pus TB +ve	Cavitation and sequestration post-laterally involving extra-capsular femoral neck
16	M	75	L	Persistent sinus formation since childhood. Several unsuccessful operations	Right hip	Before	Healed 3 years	—	—
17	M	47	L	Recurrent sinus formation 38 years: 2 unsuccessful operations	Lung	Before	Healed 3½ years	—	Extensive cavitation post-laterally involving extra-capsular femoral neck. Massive sequestration
18	M	28	L	Discomfort 7 years. "Cyst" excised 5 years. Abscess incised: sinus 3 months	—	—	Healed 4¾ years	Pus TB +ve	Grossly thickened bursa and slight bone destruction
19	F	19	R	Sinus 14 months	—	—	Healed 3½ years	Histology TB +ve	—

TABLE I—*continued*

Infection of bursa and trochanter—*continued*									
Case number	Sex	Age (*years*)	Side	History	Other lesions		Result	Confirmatory findings	Comment
					Site	Time relationship			
20	M	26	L	Mass explored 9 months before: sinus 3 months	—	—	Healed 3¾ years	Histology TB +ve. Culture TB +ve	Re-explored after 3 years for localised tenderness and thickening. Small active focus in remnant of bursa due to inadequate excision at primary operation
21	F	32	L	Sinus 14 years before; healed with conservative treatment. Recurred 4 months	—	—	Healed 5 years	—	—
22	M	33	R	Sinus 4 years before: healed on conservative treatment: recurred 5 months	—	—	Healed 6½ years	—	—
23	F	14	R	Concomitant lesions of right trochanter and hip 6 years before. Healed on 21 months' conservative treatment. Sinus 3 months			Healed 3 years. Sinus recurred	—	Extensive ramifications of sinus around hip joint. Primary lesion uncertain
24	M	28	L	Sinus 3 years before. Excision of trochanter 2 years before. Involvement of hip and subluxation within 2 months of operation			Healed 7 months after operation. Healed 7 years	—	Heavy secondary infection. Eventual bony fusion of hip

"gravitational abscesses" from tuberculosis of the lumbar spine. Thirty-two of his thirty-six patients had other tuberculous lesions before the bursal infection. Seven out of eight patients who had previously had tuberculosis of the spine had gravitational abscesses on the same side. Lampe (1953) reported seventeen cases. All the patients had abscesses and sixteen had sinuses and thirteen had past and present evidence of tuberculosis elsewhere. He believed that the lesion probably started in the bony trochanter because of the presence of encysted foci in nine cases. He obtained good results in thirteen cases and believed that radical surgery combined with antibiotics was the treatment of choice. Lindahl (1952) reported thirty-five cases of trochanteric tuberculosis. Radiographic changes were noted in twenty-five. He classified his results under four headings: eleven patients were treated by immobilisation in plaster and minor surgery (group 1); twelve by radical incision with primary suture (group 2); ten patients by radical excision and antibiotics (group 3); and two by antibiotics alone (group 4). Reactivation occurred in four cases in the first and second groups, but in none in the third or fourth group; the follow-up periods of these two latter groups were, however, short. McNeur and Pritchard (1955) reported thirty-eight cases of trochanteric tuberculosis treated at the Royal National Orthopaedic Hospital during twenty-five years. Their impressions were that the osteitis was of considerable duration and possibly preceded the bursal involvement.

TABLE II

CLINICAL DETAILS IN FOURTEEN CASES OF TUBERCULOSIS OF THE TROCHANTERIC BURSA AND GREATER TROCHANTER TREATED OTHERWISE THAN BY THE "STANDARD" METHOD DESCRIBED IN THE TEXT

Case number	Sex	Age (*years*)	Side	History	Other lesions		Treatment	Comment
					Site	Time relationship		
25	M	42	R	Erosion of trochanter noted 23 years before. Large abscess several months	Right ischio-pubic ramus	Concomitant	Excision of abscess; excision of trochanter; chemotherapy	Involvement of hip 15 months later
26	F	37	L	Pain 5 years. Sinus 4 years. Total excision of trochanter 2½ years. Involvement of hip within 1 month	—	—	Drainage of large cavity in trochanter and upper femoral region; penicillin	Heavy secondary infection. Discharge much less but sinuses persisted 5 years. Healed 4½ years. Eventual bony fusion at hip joint
27	M	48	R	Recurrent sinus formation 20 years. 2 unsuccessful operations	—	—	Removal of sequestrum; no chemotherapy	Sinus persisted 3 years. Healed 6 years
28	M	13	R	Pain and stiffness left hip 3 years. 13 months' conservative treatment both lesions. Sinus formation 7 months	Pubis	Concomitant	Total excision of trochanter	Healed 2 years. No later examination
29	M	23	L	Recurrent sinuses 6 years: 2 unsuccessful operations	—	—	Sequestrectomy	Sinuses healed, but involvement left hip 5 years later
30	M	49	R	Recurrent abscesses and sinuses 26 years: 1 unsuccessful operation	—	—	Curettage and chemotherapy	Not healed: further surgery refused by patient
31	M	57	L	Sinus for many years	Lung. Spine	Before	Curettage and chemotherapy	Healed 17 months. Sinus re-formed. Further surgery refused
32	F	65	R	Abscesses and sinuses 33 years: healed after operation. Abscess 2½ years	Right elbow	3 years before	Curettage and chemotherapy	Healed 3 years. Patient died from unspecified illness
33	F	46	L	Recurrent sinuses 33 years. Several periods of conservative treatment: 1 unsuccessful operation	—	—	Chemotherapy only; surgery refused	Failed to heal
34	M	28	R	Abscesses and sinuses 4 years. Healed for 1 year with conservative treatment: sinus recurred 2 months	—	—	Sequestrectomy	Failed to heal. Follow-up only 1 year

TABLE II—*continued*

Case number	Sex	Age (*years*)	Side	History	Other lesions		Treatment	Comment
					Site	Time relationship		
35	M	23	R	Recurrent sinuses 1 year: 4 periods of conservative treatment; several minor operations	—	—	Rest in bed 2 months	Failed to heal
36	M	47	L	Fall on trochanter 5 months before. Sinus 3 months	—	—	Healed on admission. No treatment	Healed 2½ years
37	M	56	L	Abscess 16 years before. Responded to conservative treatment plus several operations	Lung. Spine	Before	No treatment	Healed 15 years
38	M	11	R	Pain right hip after kick 11 months before	—	—	Conservative treatment for lesion of trochanter 4 years. No abscess or sinus	Re-formation of trochanteric epiphysis with coxa valga and ½-inch lengthening. The only example of a primary endosteal process. Healed at 9 months. No later examination

Twenty-five of the thirty-eight patients had tuberculosis elsewhere—this preceded the trochanteric lesion in seventeen, was concomitant with it in five, and occurred later in three. The results obtained in the earlier cases treated by immobilisation and minor surgical procedures were disappointing, and they suggested that early disease might respond favourably to aspiration of abscesses, combined with antibiotic therapy.

CLINICAL MATERIAL AND METHODS OF TREATMENT

The standard treatment adopted for the eighteen patients of whom details are supplied in Table I was as follows. Chemotherapy was begun on the morning of operation or some days before. All patients received streptomycin, usually combined with para-amino-salicylic acid (PAS) or izo-nicotinic acid hydrazide (INH) or both. At operation a liberal exposure of the greater trochanter was obtained by a long vertical incision of the fascia lata. It was often found necessary to use a transverse incision as well to excise an extensively diseased and adherent bursa. A thorough inspection of the front and back of the trochanter, as well as the extra-capsular part of the femoral neck behind, can also be carried out through this exposure. Total excision of the greater trochanter was regarded as unnecessary and dangerous, and intact bone was not removed except when necessary to drain an infected deep track in bone. After a thorough debridement the entire area was dusted with streptomycin and the wound closed in layers. Nothing more than a firm hip spica dressing was applied until the removal of sutures two weeks later. Chemotherapy was continued for six to twelve weeks and thereafter full activity was allowed. Illustrative radiographs are shown in Figures 12 to 19.

Details of the remaining fourteen patients, twelve of whom received some other form of treatment, are given in Table II. The follow-up in many of these cases has been incomplete, but enough information is available to show that the results have been unsatisfactory. The results in the whole series are shown in Table III.

FIG. 12

FIG. 13

Case 15. Figure 12—Radiograph with the hip in full lateral rotation. Figure 13—Radiograph with the hip in full medial rotation. (The erosion extended to the extra-capsular part of the femoral neck.

FIG. 14

FIG. 15

Case 17. Figure 14—Massive sequestrum in a large cavity on postero-lateral aspect of the trochanter, with smaller sequestrum in the soft tissues above. Figure 15—Appearance after operation. (The erosion extended to the extra-capsular part of the femoral neck.)

FIG. 16 FIG. 17

Case 24. Figure 16—Radiograph showing early involvement of the hip joint after excision of the greater trochanter, with several small sequestra in the soft tissues. Figure 17—One year later. Further destruction of the femoral head and subluxation of the joint.

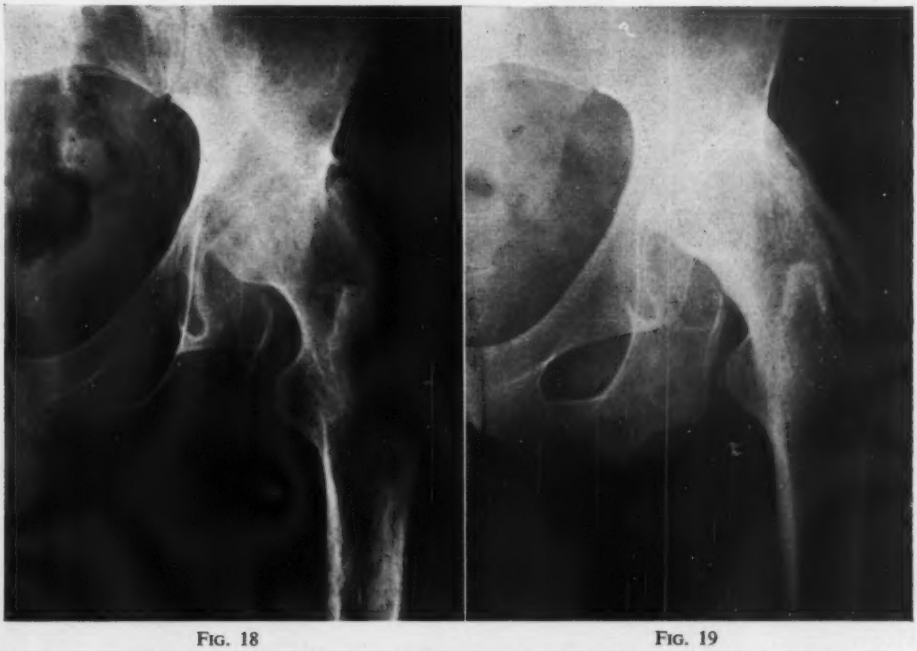

FIG. 18 FIG. 19

Case 24. Figure 18—A year later still (four months after operation). Reossification is taking place. Figure 19—Bony fusion two years after operation.

COMMENT

Case 7 was the only one in which there was early bursal involvement. The bursal sac was thin walled, contained a little serous fluid and was excised without difficulty. The remaining seven patients in the group with bursal infection alone showed marked chronic changes, especially Case 11. When bone destruction was present it was unusual to find a grossly diseased bursa. Case 18 was an exception, but in that case the erosion was slight and probably early.

Bone destruction was most marked on the postero-lateral aspect of the trochanter, and in Cases 15 and 17 there was erosion of the extra-capsular part of the femoral neck (Fig. 14). It was thought that the disease might spread to the hip joint by this route.

The patient in Case 23, who six years before had had a concomitant lesion of the right hip and trochanter, made an excellent recovery from the hip disease and retained full movement. The subsequent excision was not regarded as entirely adequate because the sinus track ramified extensively about the hip. It was not established which lesion was the source of the suppuration.

In Cases 24 (Fig. 16) and 26 the hip joint was already involved when the patients were admitted, and total excision of the trochanter had been performed previously without chemotherapy. Case 25 showed involvement of the hip joint fifteen months after excision of the trochanter and chemotherapy. Spread of the disease from a concomitant lesion in the ischio-pubic ramus could not, however, be ruled out in this case.

TABLE III

RESULTS IN THIRTY CASES OF TUBERCULOSIS OF THE GREATER TROCHANTER

Method of treatment	Number of patients	Results		
		Excellent	Good	Failed
Chemotherapy and excision of bursa and diseased bone	18	15	2*	1
Other methods	12	2	3	7

* One patient required further exploration.

(*Excellent*=First intention and permanent healing with no further symptoms. *Good*=Decreased suppuration and later permanent healing. *Failed*=Persistent sinuses, later sinus formation or spread of disease to hip joint.)

In Case 29 the lesion healed after a sequestrectomy without chemotherapy. The hip joint was involved five years later.

Case 38 was the only one in which the primary lesion was thought to be osseous. Central cavitation was present, and complete re-formation of the trochanter took place under a prolonged course of conservative treatment. There was no abscess or sinus formation throughout, but the patient had some coxa valga with half an inch of lengthening. The etiology was not proved and the follow-up was of only nine months.

CONCLUSIONS

1. The disease is very chronic and advice is sought for superficial abscess of sinus formation. While the disease remains deep to the fascia lata it is often silent or causes only minor symptoms. Time might profitably be spent in palpating the trochanteric region in the after-care of all tuberculous patients, as the thickened bursa is easily recognisable.
2. The evidence is overwhelmingly in favour of spread of the disease from bursa to bone. Eight cases out of the eighteen reviewed in Table I showed bursal involvement alone: in the remaining ten cases every degree of bone destruction from the slightest surface erosion to extensive cavitation was noted.
3. The disease probably spreads as a surface infection, although in a few cases the presence of large sequestra suggests an infection in depth at the same time. In all patients with bone

destruction the area of disease was well demarcated and a uniform hard surface of bone presented after removal of sequestra and tuberculous debris. In view of this, excision of the trochanter appears to be superfluous and possibly dangerous, even in the presence of antibiotics.

4. Radiography is an unreliable guide; decalcified sequestra and debris are often not shown. Some useful information may, however, be gained by radiographs in full medial and lateral rotation (Figs. 12 and 13).

5. In the absence of gross secondary infection the amount of discharge may be a rough but useful guide to the amount of dead tissue present.

SUMMARY

The results obtained from combined chemotherapy and radical surgery in eighteen patients with trochanteric tuberculosis have been satisfactory so far, although the period of observation is still short. The results compare very favourably with those of all other methods of treatment.

I should like to express my grateful thanks to Mr G. P. Arden, Mr W. Herschell, Mr Roy H. Maudsley and Mr F. G. Ward for their helpful suggestions in preparing this paper and for their permission to publish Cases 14, 19, 25 and 33; and to Mr Charles Gray for his permission to publish Case 6.

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OSTEOPOROSIS OF UNKNOWN CAUSE IN YOUNGER PEOPLE

Idiopathic Osteoporosis*

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Generalised osteoporosis (with the brunt falling on the spine) is gradually gaining long overdue recognition as a common disease (Meulengracht 1939; Albright, Bloomberg and Smith 1940; Black, Ghormley and Camp 1941; Albright 1947; Burrows and Graham 1947; Jackson 1955). Cooke made this syndrome his subject for the recent (1955) Lumleian Lectures. It is, of course, fundamental first of all to distinguish osteoporosis from other causes of generalised demineralisation of bone, such as osteomalacia and osteitis fibrosa (hyperparathyroidism), which are much less common (Albright 1947, Cooke 1955).

In osteomalacia the bone proteinous matrix is normal—actually excessive osteoid is formed—and there are biochemical changes in the serum, which has a low mineral content and high alkaline phosphatase. The commonest cause in a well fed community is steatorrhoea.

In osteitis fibrosa there is an excessive breakdown of bone trabeculae, excessive numbers of osteoclasts appear, and again there are distinctive biochemical changes in the serum. It is caused by hyperparathyroidism or accompanies the acidosis of chronic renal insufficiency (secondary hyperparathyroidism).

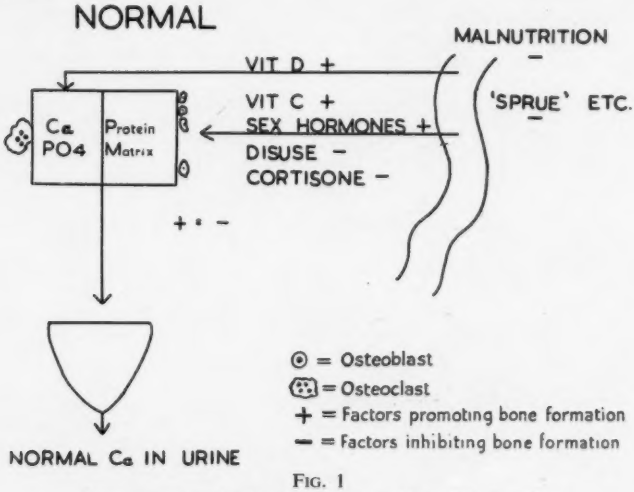

FIG. 1

The normal maintenance of bone matrix and bone salt.

In contrast, osteoporosis is a protein atrophy of bone matrix. Despite a sufficiency of minerals and normal rates of destruction, bone is laid down more slowly than it is taken away: the osteoblasts do not form enough matrix, and minerals cannot be deposited on what is not there. Osteoporosis, being, in fact, a true bony trabecular atrophy, shows no qualitative changes in the histological picture and no abnormalities in the serum chemistry (with rare exceptions of extremely rapid demineralisation, when the serum calcium may rise somewhat).

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While the porosis is developing or progressing, the normal dissolution of bone with decreased formation leads to a net loss of calcium and, presumably, phosphorus and protein from the skeleton and from the body as a whole. So much phosphorus and nitrogen come from the rest of the body that the osseous contribution of these elements to the urine is indeterminable. On the other hand, since nearly all the body's calcium normally resides in bone, any excessive loss therefrom is readily indicated by a raised urinary calcium (Figs. 1 and 2). Thus, although

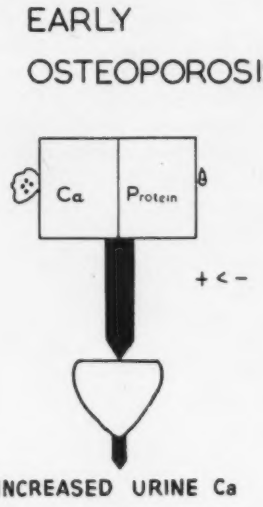

FIG. 2

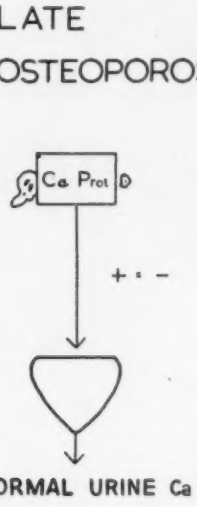

FIG. 3

Figure 2—Excreted calcium is in excess of that deposited. This diagram is really an oversimplification, showing the *net* effect—actually there is no increase in calcium release from bone, but the skeleton nevertheless loses calcium because of reduced deposition. Note the single mingy osteoblast, indicating a defect in matrix formation. (Symbols as in Figure 1.) Figure 3—Equilibrium is reattained, but with reduced bone mass. The "single" osteoblast is sufficient to maintain osteogenesis on this scale. (Symbols as in Figure 1.)

calcium metabolism is not primarily concerned in the pathogenesis of osteoporosis, we use the urinary calcium level as our yardstick of the activity of the process (we should, more accurately, assess the state of the "calcium balance"). In most cases of osteoporosis the drain of the minerals from the bones is slow, and it requires several years in negative calcium balance before symptoms arise or radiological rarefaction becomes evident (Meulengracht 1939). Gradually, then, the active bone mass diminishes until, apparently, the anabolic processes laying down bone are sufficient to balance the destructive ones, and a new calcium equilibrium is reached with respect to this small bone mass (Fig. 3). This seems to explain the frequent finding of an unusually low urinary calcium output in old men with osteoporosis, who at this stage are often free from symptoms.

Since the pioneer work of Albright and others (1940, 1947, 1948) the etiology of most types of osteoporosis is fairly well recognised. In Cushing's syndrome, whether natural or artificially produced, it is caused by the excessive glucocorticoids which actually inhibit bone matrix formation (as part of their general protein anti-anabolic activity). Immobilisation deprives the bones of the mechanical stresses and strains that seem necessary for the normal stimulation of the osteoblasts. Starvation deprives the whole body of protein (though osteomalacia is a frequent concomitant from lack of minerals and vitamin D). In old age osteoporosis may to some extent be part of general tissue atrophy (Fourman 1955), but it does seem more specifically

related to lack of oestrogens in the post-menopausal state in women, oestrogens being necessary for good function of the osteoblasts. It seems less common in men, produces fewer symptoms, and occurs at an older age (" senile osteoporosis "): perhaps in them it is related to a deficiency of androgens. Certainly hypogonadal and hypopituitary males may develop porosis, as may persons suffering from gonadal dysgenesis (formerly called " ovarian agenesis "). Other known causes include vitamin C deficiency, acromegaly, and hyperthyroidism (though more than osteoporosis occurs here (Follis 1953)). It has been reported also in hypothyroidism in children and adults (Fourman 1955), and we have seen it in hypoparathyroidism. Its occurrence in chronic liver disease is somewhat doubtful, though one can imagine the persistent low serum albumin being related to it. Hernberg (1952) asserted that it has a high incidence in diabetes, but this must also be considered non-proven. We believe that osteoporosis (not confined to the relatively fixed thoracic spine) is common in chronic emphysema, and also appears in rheumatoid arthritis even when the back does not seem to be involved in the arthritic process, but these ideas await further work. The possible connection of osteoporosis with pregnancy is considered later. Finally, it is not illogical to consider osteogenesis imperfecta as " congenital osteoporosis."

" Idiopathic osteoporosis "—Despite all these known or possible explanations for osteoporosis there remains a group of cases in which the cause is entirely unknown. Affected people are comparatively young and seem to exhibit no common etiological denominator. Although the condition is rather uncommon, yet, since it occurs in the younger wage-earning population and is apt to be severe and disabling, it is nevertheless important.

We call this syndrome " idiopathic osteoporosis," and it must be admitted from the outset that this paper does not produce any good theory of etiology or line of therapy. The reason for my writing it is that I can now present an analysis of thirty-eight cases of what is generally considered to be a rare condition. In many quarters it is even unknown, and consequently unrecognised.

LITERATURE ON IDIOPATHIC OSTEOPOROSIS

There is very little literature on this subject. This may be partly because of the lack of distinction of this group from the more common osteoporotic conditions of older people and partly because of our present paucity of knowledge regarding etiology and effective treatment.

Meulengracht (1939) included three men in their forties in his series of osteoporotics. They seem to have been typical of the " idiopathic " group. He treated them with vitamin D and calcium and claimed that two improved, whereas one got worse. Burrows and Graham (1947), under the title of " Spinal osteoporosis, cause unknown," included people of both sexes and all ages, six of the males being under fifty-five. They noted one young woman in whom the condition was seemingly " precipitated " by pregnancy. They took pains to distinguish the condition as a whole from osteomalacia and reproduced some excellent radiographs and histological preparations, but found no positive etiological factor. Most of their cases would now be considered " post-menopausal " or " senile."

Albright and his collaborators (1944, 1948) mentioned a few cases of osteoporosis in young people, and believed them to constitute a special group. They did not, however, consider this aspect of the subject in detail, reporting rather on the effects and fate of intravenously administered human serum albumin in these particular patients.

Anderson (1954) reported the case of a man aged thirty with this condition, and claimed symptomatic improvement and calcium retention on treatment with intravenous plasma. Nordin and Roper (1955) described four young women who developed osteoporosis, apparently associated with pregnancy; and these cases, despite the strictures of Brailsford (1955) which followed their paper, seem to be genuine enough. They remarked upon a similar case reported by Gössler (1953), and made the interesting suggestion that the increased adrenocortical activity in pregnancy might be responsible, as it is in Cushing's syndrome. Osteomalacia was definitely ruled out.

Dent (1955) reported four patients, one man and three women. In only one of the three women did the symptoms closely follow pregnancy, and one had never been pregnant at all. The women did not improve with plasma infusions.

CLINICAL FEATURES

The symptoms and signs are virtually those of any variety of spinal osteoporosis, as described elsewhere (Meulengracht 1939, Black *et al.* 1941, Burrows and Graham 1947, Dent 1955).

Pain is usually the outstanding feature, but it may be completely absent, even in advanced porosis. Often the mode of onset is of agonising pain localised in the spine, and related to an acute compression fracture of the vertebral body. A known injury, usually of insufficient severity to have fractured a normal spine, may or may not have produced this acute damage: sometimes merely the strain of bending to pick up an object is sufficient. Activities that have been responsible for acute damage to patients in the present series include: slipping on concrete steps (three cases), " lifting a car " (two cases), diving, swinging a golf club, sneezing (two cases), convulsive shock therapy (two cases), lumbar puncture, playing " bok-bok " (a South African jumping-over-backs game), playing soft-ball, jumping from a train, falling against a barrel, and falling from a horse, from a bus and from a lavatory seat. When there is a history of trauma it is, of course, most important to take note that the uninjured part of the spine is porotic, otherwise the diagnosis may be missed.

Alternatively, the onset may be indefinite, with vague aching pains in the back, becoming more severe. Stabbing pains both locally and of girdle distribution may occur, but sciatic radiation seems uncommon, despite the frequent presence of compression of the lower lumbar vertebrae. Meulengracht (1939) described one man in whom attacks of pain resembled angina pectoris and were felt also in the left shoulder and down the left arm. The spontaneous pain is usually intermittent, but may be incapacitating for a variable time, even for months; and in intervals of comparative freedom, any sudden movement of the spine, sneezing or lifting may precipitate further pain.

The pain is usually thoracic or lumbar, sometimes cervical. In one case in this series it was felt mainly in the sternum and in another case in the pelvis. Some patients complain of pain in sites remote from the spine, especially the feet (the bones of which are then found to be porotic) and sometimes in the proximal limb joints, without any evidence of contemporary or subsequent arthritis.

Several times I have been struck with the story of loss of weight at the onset of the complaint, general listlessness and tired feeling, easy fatigue and loss of energy. The patients may look unwell, and their sedimentation rate may be raised. (This was so in six out of eighteen patients tested at the outset.) Careful examination fails to reveal other disease, and the general health and sedimentation rate return to normal later. Perhaps these symptoms may be explained by the constant pain, but possibly they may be part of the syndrome itself.

The patient may seek advice for symptoms caused by fractures of bones other than the vertebrae. A native man (the only Bantu in the present series) complained of pain in a lump on the sternum. A fractured sternum was found. The neck of the femur is particularly liable to fracture in osteoporosis from any cause; in fact, with rare exceptions, this fracture is always a pathological one, associated with porosis. Fracture of the ribs, clavicle, pelvis and neck of humerus have also been presenting features or complications. On attempting to obtain marrow it may be observed that the sternum offers little resistance to the needle.

Associated with the back pain is an immobility which is usually complete during the acute phases. In these people who are pain-free it is interesting to observe good spinal flexion, even with multiple compression fractures. It seems even as if their ability to touch their toes is better than normal; perhaps the tendency to wedge-compression of the vertebral bodies mechanically facilitates flexion.

Secondary anatomical effects, depending on the extent of spinal compression, include loss of height, a permanent stooping posture, approximation of the lower ribs to the iliac crests and redundant skin folds across the abdomen. Despite the prevalence of girdle pain, presumably from nerve root irritation, objective neurological features of spinal or root compression do not occur.

RADIOLOGICAL FEATURES

The radiographic changes have been well described and illustrated before (Burrows and Graham 1947, Albright and Reifenstein 1948) in other varieties of osteoporosis and, in general, the appearances are the same. In the vertebral bodies there are two types of change (Fig. 4).

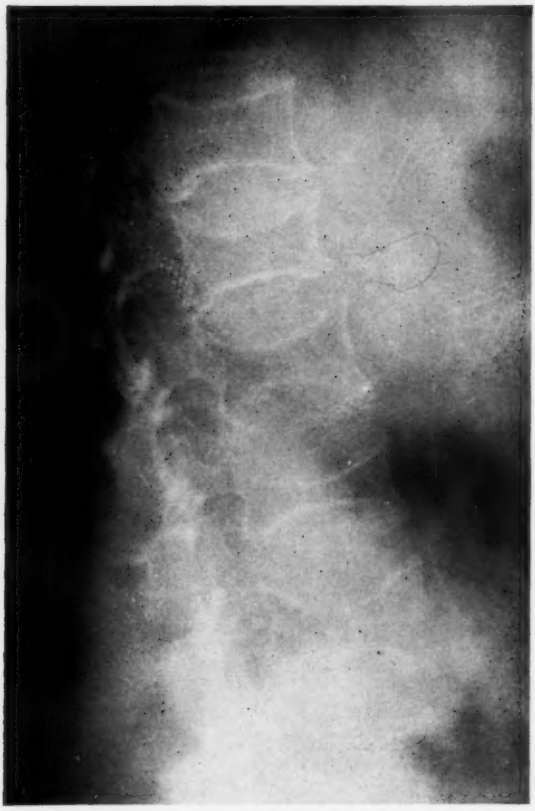

FIG. 4

Typical severe osteoporosis (Case 2). Vertebral bodies are homogeneously translucent, with irregular but gross compression of all bodies by the adjacent expanded discs. (Dr Albright's case.)

Internal structure—A generally homogeneous appearance with loss of trabeculation and an increase of translucency may be seen; alternatively the trabeculations (especially the vertical ones) may seem to stand out more plainly than usual (Kesson, Morris and McCutcheon 1947; Moldawer 1955) as if " the branches having been removed, the trunks are more prominent " (metaphor used by Albright). An important sign is the " pencilling " of the cortex of the vertebral bodies: the cortices are thin, but stand out against the more porotic medullae. Nevertheless it is usually difficult and dangerous to diagnose osteoporosis on these features

alone, because enormous variations may depend upon differences of technique and upon the thickness of the overlying soft tissues.

Changes in shape—These include ballooning of discs into the vertebrae producing biconcavity of the bodies, more localised disc herniations (Schmorl's nodes), small breaches in continuity of the vertebral plates, anterior wedging, and compression of varying degree of the whole vertebrae. Every case in the present series has shown one or more of these changes, which are characteristically remarkably irregularly disposed throughout the thoracic and lumbar regions of the spine.

After wedging or compression has been present for a long time osteophytic outgrowths occur from the vertebral bodies in the region of the cartilaginous plates, often from contiguous vertebrae. Anterior vertebral osteophytes, which may actually form bridges between adjacent

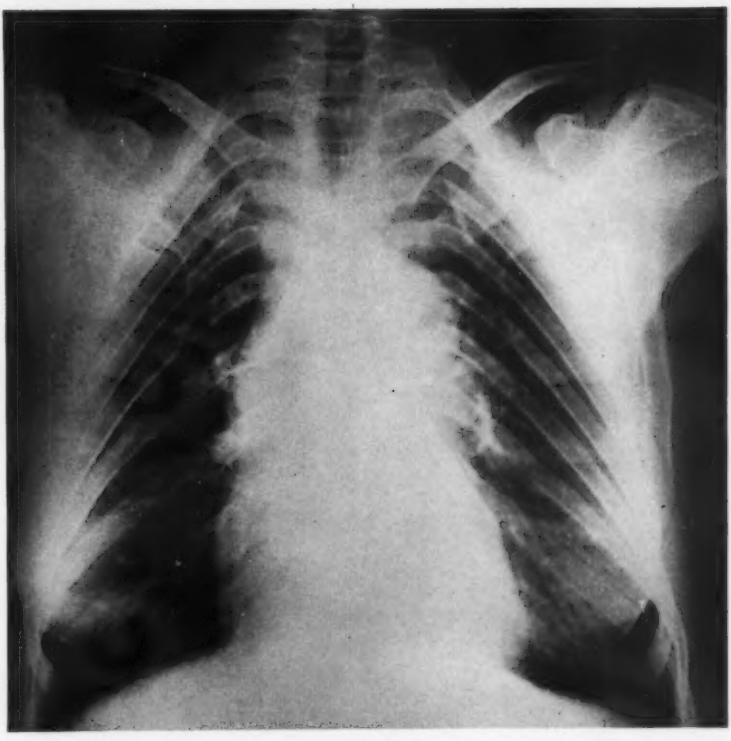

FIG. 5
Typical "isosceles triangle" shaped chest in a woman with idiopathic osteoporosis. (Case 37).

bodies, are, of course, well known and commonly seen in spinal radiographs. They are frequently ascribed to "osteoarthritis," but they are not related to synovial joints and in fact are usually secondary to a prior vertebral disorder either of bone or cartilage. Their presence, therefore, must make one look carefully to discover the underlying lesion. If this lesion is a compression fracture of osteoporosis they further indicate that this fracture must be of some age.

As in all forms of osteoporosis, other proximal bones are involved, but to a lesser degree. As mentioned above, fractures of these bones may be seen, and they do not unite easily, forming but little callus. In severe cases the long bones, hands, feet and skull may also become porotic. To be sure of porosis of the skull is difficult, but in this series there were two very definite

cases, closely resembling myelomatosis radiographically. This condition has been ruled out by the length of time during which the patients have been observed, and in one instance by biopsy of the skull vault. In a few other radiographs demineralisation of the clinoid processes has been seen. (This is a well known feature of osteoporosis, usually commented upon in Cushing's syndrome.)

The women, particularly, seem to develop deformities of the thoracic cage. Its capacity is diminished, the ribs slope more acutely downwards, the rib spaces tend to disappear in the antero-posterior view, and the general shape is that of an isosceles triangle (Fig. 5).

BIOCHEMICAL FINDINGS

As in other varieties of osteoporosis, there are no changes in the serum chemistry. It has been claimed that the serum phosphorus tends to be on the high side of normal in the post-menopausal group (Albright and Reifenstein 1948), but this has not been found in the "idiopathic" cases in younger patients. In the present series the mean serum phosphorus was 3·3 milligrams per 100 millilitres with a range of 2·1 to 4·3. The serum alkaline phosphatase has seemed to be on the low side in a few cases, and this is discussed below.

The characteristic finding in the urine is a high calcium output. On a normal diet, in which no milk by the glass or calcium tablets are being taken, we consider 150–200 milligrams a day to be suspicious and over 200 to be definitely high. Sixteen patients out of twenty-seven who were tested when first seen were found to have a high or suspiciously high urine calcium output; in eleven it was within normal range. It may be presumed that in these eleven patients it had been high previously, and high for a long time, but that the "porotic process" had ceased or was at least quiescent when they were seen.

The plasma albumin was over 4 grammes per cent in every male case; in two of the women there was a reversed albumin/globulin ratio; and flocculation tests were abnormal in one.

A glucose tolerance curve was performed in eight patients; in three the figures were rather high at two to two and a half hours; one other patient was a known diabetic.

The output of 17-ketosteroids was within normal range in all but two men and one woman out of seventeen patients in whom it was measured. In these three it was low. The mean 17-ketosteroid level of the whole group was 20 per cent below the normal mean (according to Hamburger's chart), each case being separately estimated. This does not seem to be a significant change.

In seven cases the gonadotropin output in the urine (follicle-stimulating hormone, F.S.H.) was measured by the immature mouse uterus test and found to be unusually high in one case, and low in two. Blood count and bone marrow were not abnormal in any case.

ASSOCIATED CONDITIONS

The possible existence of liver disease, epilepsy, diabetes, dysthyroidism, malabsorption and malnutrition was particularly considered. In six cases there was a history of a fairly large intake of alcohol, but no patient had obvious liver disease, and in only one was any "dysfunction" found by flocculation test. One epileptic is included, and two patients who had received shock therapy. Fractured vertebrae were probably related to convulsions in each case.

No serious gastro-intestinal disease was present in any patient; steatorrhoea was not found (fat balance was performed in four cases). Free hydrochloric acid in gastric juice was present in all tested. Four patients had duodenal ulcer.

One patient developed diabetes. One patient who was later discovered to be thyrotoxic has been excluded from the series. In two cases the possibility of scurvy was considered. In one, the only pure native, scurvy was probably present for a short time before he was seen; in the other it was very doubtful. One woman developed myasthenia gravis after osteoporosis; another also had familial metaphysial dysplasia; a third had a patent ductus arteriosus.

HISTOLOGY OF THE BONE

Since osteoporosis is an atrophy of the proteinous bone matrix, there are no striking histological changes. No necropsy material is available. Biopsy of tibia, skull or rib has been performed in eight cases of the present series and in each the finding has been similar. As in other varieties of osteoporosis (Burrows and Graham 1947, Cooke 1955, Sissons 1955) there are: 1) thinning and decrease in the number of trabeculae; and 2) a small number of osteoblasts. These quantitative changes can be demonstrated with certainty only by comparison with a normal control.

Biopsy is probably of greatest value in ruling out other diseases (such as myelomatosis), but it cannot be expected to distinguish osteogenesis imperfecta, because this is itself an osteoblast-deficient osteoporosis.

THE PRESENT SERIES

To the only twelve cases found to be separately described in any detail in the literature the present series adds thirty-eight, twenty-seven in men and eleven in women. I have thought it reasonable to include in this group men who had recognisable porosis before the age of fifty-five, or women before the menopause.*

The source of origin of the members of this series is various. Fifteen of the men and eight of the women were patients of Dr Fuller Albright at the Massachusetts General Hospital or were referred to him. About half of these were seen by the author; of the rest, the protocols, radiographs and other available documents were carefully examined, and all were written to. Nine men and two women were seen and investigated personally at Groote Schuur Hospital, Cape Town. Two men were patients of Dr I. J. Grek of Johannesburg, to whom I am indebted for the protocols and radiographs, and one man was seen at the Maclean Hospital, Harvard, by courtesy of Dr Mark Altschule.

All the Americans were white; of the South Africans, two men and one woman were coloured; one man was pure Bantu (" native ").

The mean age at which the condition began, judging from the onset of symptoms, was twenty-seven for the women and forty-one for the men, with a range of twenty-three to fifty-five in the men and twenty to forty in the women.

All the cases are briefly summarised in Table I; some are described in greater detail below.

ILLUSTRATIVE CASES IN MEN

Case 24†—White, present age sixty-three, age at onset of symptoms fifty (Fig. 6). A hairdresser by occupation, always a very active man, a gymnast when young. Onset was sudden, while bending to pick up a piece of soap in the bathroom. The pain was in the lower lumbar region, very severe and completely incapacitating. The patient was losing weight and feeling generally tired at that time. He was immobilised for two or three months at the start (never since) and suffered severe intermittent relapses at about yearly intervals up till three years ago, with lower lumbar and girdle pain. He has had no pain for three years; just feels somewhat " stiff." He has used a spinal brace since 1946, but is now discarding it for short periods.

FIG. 6
Case 24—Photograph of the patient showing thoracic kyphosis and compression of abdomen between thorax and pelvis.

* It must be realised that in fact the disorder of metabolism must have been present for several years (probably at least five) before the onset of symptoms or signs, in which " pre-osteoporotic " phase the bones were continually losing substance. A man of fifty should not be senile, nor grossly lacking in sex hormones.

† Patient seen in early stages and carefully followed by Mr Arthur Helfet, to whom I am much indebted.

TABLE I

CLINICAL DETAILS IN THIRTY-EIGHT CASES OF IDIOPATHIC OSTEOPOROSIS

Case number	Age at onset (*years*)	Type of onset	Severity	Pain elsewhere than in spine	E.S.R. (*mm.*)	Sexual function	17-ketosteroids (*mg./day*)	Urine calcium when first seen (*mg./day*)	Radiographic distribution of porosis	Progress
					MALES					
1	31	Fall	++	—	26	Normal	—	107	Spine (compression)	Intermittent attacks
2	34	Gradual	+++	Feet	6	Normal	7–14·5	300–600	Spine; feet; skull	Improved gradually
3	34	Gradual	+	—	—	Reduced	8–13	—	Spine	Improved
4	42	Gradual	+	—	3	Normal	9·5	135	Spine	—
5	43	Gradual	++	Heel	—	Normal	8–10	300	Spine; rib (fractures)	Worsening
6	37	Gradual	+	Foot and shoulders	8	Normal	5	400	Spine (collapse); ribs (fractures)	Worsening
7	55	Sudden	+++	—	—	Testicular atrophy	—	104	Spine (collapse)	Improving
8	55	Sudden	+++	—	—	Reduced	6	30	Spine; skull	Improving
9	36	Abrupt	++	—	14	Normal	14	200–400	Spine	Improving
10	24	Abrupt	+	—	—	—	—	245	Spine; neck of femur (fracture)	Slowly progressed
11	53	Gradual	+++	Legs	6	—	—	250	Spine	Improved
12	52	Sudden	++	—	48	Normal	12	300	Spine	—
13	30	Sudden	+	—	—	Normal	10	240	Spine	Progressing
14	Unknown	—	0	—	—	Depressed	—	—	Spine	Pain-free (schizophrenic)
15	36	Gradual	+++	—	32	—	—	—	Spine; sternum (fracture)	Stationary
16	23	Rapid	++	—	2	—	—	200	Spine	Intermittent
17	50	Gradual	+	All big joints	41	Normal	—	30–60	Spine	Improved
18	37	Gradual	++	Knees and ankles	20–30	Testicular atrophy	4	Low	Spine	Stationary
19	47	Sudden	++	Thigh	—	Normal	—	300	Spine	Improved
20	37	Sudden	++	—	8	Normal	—	400	Spine	Improved
21	50	Sudden	+++	—	39	Testicular atrophy	7·6	150–300	Spine	—
22	37	Sudden	+	—	4	Normal	—	150	Spine	—
23	Unknown	Gradual	+	—	4	Impotent	—	300+	Spine; ribs (fractures)	Recent pain

TABLE I—(*continued*)

23	Unknown	Gradual	+	—	4	Impotent	—	300+	Spine; ribs (fractures)	Recent pain

TABLE I—(*continued*)

CLINICAL DETAILS IN THIRTY-EIGHT CASES OF IDIOPATHIC OSTEOPOROSIS

Case number	Age at onset (*years*)	Type of onset	Severity	Pain elsewhere than in spine	E.S.R. (*mm.*)	Sexual function	17-ketosteroids (*mg./day*)	Urine calcium when first seen (*mg./day*)	Radiographic distribution of porosis	Progress
MALES										
24	50	Sudden	+++	—	Normal	Normal	7·6	180	Spine; skull	Much improved
25	43	Gradual	+	—	3	Normal	—	—	Spine	—
26	53	Gradual	+	—	—	Normal	—	—	Spine	—
27	?51	Rapid	++	—	3	Depressed	11·2	400–600	Spine (compression); ribs (fractures)	Improved
FEMALES										
						Relation to pregnancy				
28	24	Gradual	+	—	—	Onset after delivery	—	32	Spine	—
29	22	Gradual	++	Feet	—	Never pregnant	4·7	219	Spine; long bones	Stationary
30	32	Gradual	++	—	—	Onset during lactation	—	120–140	Spine; clavicle (fracture)	Improved
31	32	Gradual	+	—	—	Onset on honeymoon	—	—	Spine; ribs; clavicle (metaphysial dysplasia)	Stationary
32	24	Gradual	+++	Feet	27	Onset during pregnancy	6–16	200–300	Spine; skull; ribs; long bones (fractures)	Worsening
33	28	Gradual	+++	Knee	—	Onset during pregnancy	4–6	32	Spine; long bones (several fractures: ? osteogenesis imperfecta)	Improved
34	20	Rapid	++	Ankles	—	Onset during pregnancy	12·3	50–150	Spine; ribs; femur; pelvis (fractures)	Improving
35	21	Gradual	+	—	—	Onset during pregnancy	5·9	186	Spine	—
36	25	Gradual	+	—	—	Onset during pregnancy	—	100–250	Spine	—
37	43	Gradual	+	—	16	Only pregnancy 25 years ago	—	220	Spine	Mild
38	24	Sudden	++	? (hysterical personality)	3	Only pregnancy 4 years ago	2·8	150–250	Spine	Improving

He had had no major accident or serious illness; never felt pain in the joints and never indulged in alcohol. He is married with two children; libido has been retained, its expression being curtailed simply by his back pain; nocturnal emission still occasionally takes place. He eats well and takes quite normal food; there is no suggestion of gastro-intestinal disorder.

He is a small, bent man (Fig. 6) with typical posture and abdominal skin fold. He has lost several inches in height. Nevertheless he can now bend to within eight inches of his toes, which is a great improvement. His chest expansion is good and his lungs are clear. There is no obvious disorder of thyroid, liver, kidneys, testes or blood.

Investigations—In 1946 the erythrocyte sedimentation rate was not raised. Haemoglobin 14 grammes per 100 millilitres. Serum calcium 9·3 and 10·7 milligrams per 100 millilitres; serum phosphorus 2·4. Serum alkaline phosphatase 2·4 and 3·9 units (Bodansky). Glucose tolerance normal. Twenty-four hours' 17-ketosteroid excretion 7·6 milligrams. Basal metabolic rate +12 per cent. Calcium balance 60–70 milligrams negative daily (two experiments).

In 1955 the twenty-four hours' urinary F.S.H. was positive at 24 mouse units.

Twenty-four hours' urine calcium (in milligrams) was as follows: 1946—171, 179 (on 400-milligram intake); 1947—150, 156 (on 800-milligram intake); 1954—119 (on 800-milligram intake).

Radiographic examination—In 1946 there was generalised spinal osteoporosis with several collapsed vertebrae and "ballooning" of the disc spaces. In 1955 the appearance was similar, with some further vertebral wedging. Secondary osteophyte formation had occurred, with calcification of the discs in the lumbar region. The feet were doubtfully affected, the pelvis grossly so. The clinoid processes of the skull were barely visible. No fractures were seen outside the vertebrae.

Treatment—Several lines of treatment were tried, but it cannot be said that any was successful, except for the brace which relieved the aching and gave a feeling of stability.

Comment—A clear case of osteoporosis in an active man, with normal sex life and no indication whatever as to the etiology of the syndrome. There is a tendency to spontaneous slow improvement.

Case 20—White, present age forty-six, age at onset thirty-seven (Fig. 7). A farmer, a big and strong man, felt a sudden sharp pain in the lumbar spine on straightening up from a bending posture. For four years he had constant aching, with periodic exacerbations demanding bed rest. He was married, with two children, and there was nothing relevant in his eating habits, alcohol consumption, sex life or past and family history.

On examination the thoraco-lumbar spine was immobile. There was tenderness over the spinous processes of L.2 and L.3, and he could not bear any jarring. He was not otherwise affected.

Investigations—In 1951 the erythrocyte sedimentation rate was 8 millimetres in the first hour. Haemoglobin 15·7 grammes per cent. Serum albumin 4·2 per cent; serum globulin 2·1 per cent; serum calcium 9·6 milligrams per 100 millilitres; serum inorganic phosphorus 3·2 milligrams per 100 millilitres; serum alkaline phosphatase 5·5 and 1·7 units (Shinowara). Test meal showed free hydrochloric acid present. The fat balance was normal. Twenty-four hours' urine calcium was 350–400 milligrams.

Radiographic examination—There was very severe porosis of the whole spine, and of the pelvis and ribs. There were multiple compression fractures in the thoracic and lumbar spine, with general biconcavity of the vertebral bodies and infractions through several vertebral plates (Fig. 8). In the skull there were numerous rounded translucencies, reminiscent of myelomatosis (Fig. 9). As pointed out by Dr L. Werbeloff, these appearances are confined to the diploë, the inner and outer tables not being eroded. This is a strong point against myeloma. The sella turcica and clinoid processes are so faint as to be barely visible. Decalcification is much less certain in the long bones and hands.

Metabolic balance studies were performed only after treatment with methyl testosterone (25 milligrams a day) and vitamin D (20,000 units a day) had been begun. On this treatment the patient was just in balance for calcium. The addition of a high protein diet (140 grammes a day) and a gramme of extra calcium as calcium gluconate produced a positive calcium balance of 300–400 milligrams a day, and this was maintained for ten weeks. The patient felt much better during this treatment, put on weight and became more active (possibly merely in consequence of natural fluctuations in the disease process).

Progress—A brace was worn at first but has recently been replaced by a surgical corset. Despite this and the treatment already mentioned, further severe attacks of pain occurred until 1953, since when his ability to bend has much improved and there has not been severe pain.

Comment—In this younger man there was again no clue to the etiology of the porosis. The medical treatment offered was empirical. Nevertheless it certainly produced a positive calcium (and presumably nitrogen) balance, at least for several weeks.

Case 21—Coloured, age fifty. This motor car sprayer slipped on a concrete floor immediately before his admission and sustained a compression fracture of the third lumbar vertebral body, with severe pain. The rest of the spine was markedly porotic, with typical alterations in vertebral contour. Other

FIG. 7

FIG. 8

Case 20. Figure 7—Photograph of patient. Note puckering of skin caused by vertebral collapse with loss of height, more marked on the right than the left. Figure 8—Lumbar spine. Porosis with irregular compression of vertebral bodies and infractions into vertebral plates.

FIG. 9

Case 20—Skull. Note the " spotty " generalised porosis.

bones were more doubtfully affected. He was accustomed to taking considerable quantities of alcohol. There was no evidence of other significant disease from the history or examination, except of the sexual system. He was married, with four children, the youngest four years old. For two years he thought his desire had lessened. Bilateral testicular biopsy revealed moderately severe atrophy with much reduced spermatogenesis, disorganisation of seminiferous epithelium, thickened basement membrane and peritubular fibrosis. Sertoli and leydig cells were not obviously abnormal.

Investigations—The erythrocyte sedimentation rate was initially 39 millimetres in the first hour (Westergren). Four weeks later it was 21 millimetres. The serum albumin was 4·2 per cent; serum globulin 3 per cent. The flocculation tests were normal. Serum calcium 11·2 milligrams per 100 millilitres; serum inorganic phosphorus 4·8 milligrams per 100 millilitres; serum alkaline phosphatase 5 and 5·4 units (Shinowara). Blood urea 30 milligrams per 100 millilitres. Twenty-four hours' 17-ketosteroid excretion was 7·6 milligrams. Twenty-four hours' urine calcium was 147 milligrams (mean) and 264 milligrams after second fracture.

Course in hospital—Rest in bed was required at first because of the severe pain. After ten days he was encouraged to get up, for fear that longer immobility might increase the porosis. Almost immediately, despite care in avoiding bending, he got a second attack of severe pain and it was found that partial compression of another vertebral body had occurred. A few weeks later he was fitted with a spinal brace, which has been used intermittently and without further mishap.

In view of the testicular histology treatment with testosterone was given, but no follow-up has been possible.

Comment—This patient taught us the importance of recognising the " acute phase " of this disease, during which fractures are more liable to occur. The periodicity of the pain, so typical of many of the histories, also suggests a phasic activity. Exacerbations of symptoms, however, do not necessarily seem to coincide with exacerbations of calcium excretion.

THE DISEASE IN WOMEN

Relationship to pregnancy—In each of Nordin and Roper's four cases in young women the onset was related in time to late pregnancy or the period of lactation. In one case there was a recurrence of symptoms with a later pregnancy. These authors noted a few similar cases in the literature (Burrows and Graham 1947, Jones 1953, Gössler 1953). However, in only one of Dent's (1955) three young women patients did the symptoms relate to pregnancy, and one had never been pregnant.

In the present series there was an apparent relation to pregnancy or lactation in seven of the eleven women. In one of the other four (Case 37) the onset of symptoms was more than fifteen years after her single pregnancy; in one (Case 31) the first symptom was caused by a compression fracture sustained during her honeymoon; one (Case 29) was never pregnant; and one (Case 38) was pregnant four years before the onset of symptoms: she complained of no pain at that time.

In two of the cases with onset related to pregnancy the symptoms recurred during subsequent pregnancies, but in another one (Case 34) they were actually relieved at this time. In one case (Case 32) a balance study showed considerable improvement in the metabolic state after the termination of pregnancy.

That pregnancy is actually the cause of the disease seems very unlikely, for the development of such severe porosis in the short space of nine months would require a very rapid calcium loss indeed. In fact if one assumes a loss of one-third of the body's kilogram of calcium, then the urine would have to contain over a gramme a day. This certainly does not occur. On the other hand the above evidence strongly suggests a relationship to pregnancy in some cases. It seems likely that pregnancy may be a contributory or precipitating factor in a person already predisposed to the disease, or, in fact, in a person who already has osteoporosis but has not yet suffered symptoms. Such an effect of pregnancy might be related to the extra demands for protein and minerals, or, more subtly, to the increased activity of the adrenal cortex which is known to occur (Nordin and Roper 1955). If the latter is an operative factor, then pregnancy is here comparable to Cushing's syndrome.

Diagnosis—Regarding the diagnosis in these women, there seems no doubt that osteomalacia,

FIG. 10

FIG. 11

Case 32. Figure 10—Lateral thoracic spine showing kyphosis, porosis, vertebral collapse and ballooning of disc spaces. There was also a fracture of the sternum. Figure 11—Feet, showing abnormal translucency of the bones, with very thin cortices. (Dr Albright's case.)

FIG. 12

Case 32—Pelvis. There is general osteoporosis, with " pencilled in " cortices, and pathological fractures of the neck and shaft of the left femur.

the cause of spinal collapse during pregnancy in certain countries, can be ruled out. As with the men, the dietary, clinical, biochemical and radiological features are all against this: the condition is not relieved by mineral and vitamin D therapy, and biopsy in four cases demonstrated normal, thin bone, without any specific features. In one case (Case 33) there was some bending of bone, as shown particularly by a tri-radiate deformity of the pelvis. The diagnosis in this case is very difficult, and although osteomalacia was excluded, osteogenesis imperfecta cannot be entirely ruled out.

Course—The course of the disease in women has been variable. Three of the women with onset related to pregnancy have been followed for several years. One is improved at the end of seven years and is doing housework; one is worse after fourteen years and just hobbles about on crutches; and the third remained much the same for ten years, at the end of which she sustained a spontaneous fracture of the femur and of the humerus.

Comparison with the condition in men—In this series the incidence of idiopathic osteoporosis has been greater in men than women (27 : 11). There are not enough well documented cases of this whole group in the literature to work out the sex incidence further, but it is noticeable that Nordin and Roper (1955) did not mention any males, and Dent (1955) quoted one out of four. In other series (Burrows and Graham 1947) the possibility of early or artificial menopause or other endocrine disturbance, for instance, does not appear to have been excluded.

However, the average age at onset in the women is much lower than in the men (twenty-seven years as against forty-one). If pregnancy and lactation are indeed causal factors, it is suggested that this added strain might precipitate symptoms at an earlier age in those already prone to osteoporosis.

Regarding skeletal changes, I get the impression that there tends to be more involvement of the thoracic cage in the women, with greater likelihood of sternal protrusion, rib space closure, immobility and diminution of vital capacity.

In general, I believe that this is the same disease in the two sexes, though with modifications by special circumstances pertaining to either sex.

ILLUSTRATIVE CASE IN WOMAN

Case 32 (Albright and Reifenstein 1948)—White. In 1938 at the age of twenty-three she jumped from a fence and has had pain in the feet since. In 1939, during first pregnancy, developed pain in back and ribs, with general weakness. In 1943 first seen by Dr Albright. Again pregnant, with back pain. She had severe spinal porosis, with multiple compression fractures (Fig. 10). All the bones were demineralised, including the feet (Fig. 11) and the skull, though the lamina dura of the teeth was intact. During the course of her illness she broke both femoral necks (Fig. 12), one tibia and several ribs.

The only biochemical abnormality was a high urine calcium. Several metabolic balance studies were carried out. Untreated, she was in negative balance. There was a distinct improvement in the balance after abortion, but a high protein diet, vitamin A and stilboestrol had no value. Repeated intravenous infusion of concentrated human serum albumin produced a nitrogen and calcium retention, improvement in symptoms and a reduction of urinary calcium to zero. Albright points out that the periods when the balance tended to be positive coincided with the highest serum protein levels. It was largely on the results of these and similar experiments that he developed his hypothesis that serum albumin is the precursor of bone matrix (Albright *et al.* 1946).

Diagnosis—Hyperthyroidism was excluded by a normal basal metabolic rate, a protein-bound-iodine of 7·5 mg., and a radio-iodine uptake of 53 per cent. Tibial biopsy showed qualitatively normal bone.

Course—The disease has been phasic, but on the whole the course has been downhill, with severe painful episodes, increasing loss of height and deformity. After fourteen years she could just hobble around on crutches, tiring easily, with grossly deformed thorax and poor vital capacity.

DIFFERENTIAL DIAGNOSIS

In old people the diagnosis of osteoporosis is comparatively easy. It is always necessary, however, to look for evidence of myelomatosis, carcinoma and thyrotoxicosis. The rarity of

the disease in younger people makes diagnosis more difficult and the criteria stringent. The following points are important. 1) Demineralisation is greatest in the spine, but the degree of damage to vertebral bodies is not uniform. 2) There is no bending or bowing of bones, and no cyst formation or localised erosion. The lamina dura of the teeth remains intact. 3) Serum chemistry is normal in all respects. 4) The urine is normal except for an inconstantly high content of calcium. 5) Prolonged immobilisation, malnutrition (including scurvy), food-faddism and steatorrhoea are absent. 6) The above criteria being satisfied, other specific diseases must be ruled out. These will now be considered.

Myelomatosis can mimic the spinal changes of osteoporosis in that occasionally the changes are diffuse. Patients with gross bone involvement would, however, be seriously ill, with high serum globulin and raised sedimentation rate. The bone marrow should be examined if any doubt remains.

Hyperparathyroidism (with osteitis fibrosa)—Except in Case 23 there was no difficulty in excluding hyperparathyroidism on the evidence, in hyperparathyroidism, of 1) renal calculi (usually); 2) the characteristic changes in the serum chemistry; and 3) radiographic features other than pure porosis (cysts, periosteal erosions, bone expansion).

In Case 23 differentiation from hyperparathyroidism was difficult because of the occurrence of multiple calcium-containing renal calculi, of alterations of the serum chemistry (calcium 11·6 milligrams per cent (highest); phosphorus 1·4 milligrams per cent (lowest)); and of a twenty-four-hour urinary calcium excretion of 300 milligrams on a low calcium intake of 130 milligrams a day. Apart from the spine, there was osteoporosis of the ribs, with several fractures, and of the skull, with translucent clinoid processes but intact lamina dura. The tibiae and hands appeared normal. There was gross calcification of the aortic and iliac arteries. Against the diagnosis of hyperparathyroidism are: 1) remission during the last four years in chemistry and renal state, with normal renal function (if parathyroid disease had been present it must have "burnt out"—a very rare happening, and probably only explicable on the basis of infarction into a parathyroid adenoma (Howard *et al.* 1953)); 2) normal alkaline phosphatase with marked bone disease; and 3) the distribution of the decalcification, the absence of cysts and the retention of the lamina dura. On the whole, I favour the diagnosis of idiopathic osteoporosis, with the unusual, but theoretically probable, complication of renal calculi.

Hyperthyroidism—Follis (1953) claimed that, histologically, a mixture of osteomalacia, osteitis fibrosa and osteoporosis may be found in thyrotoxicosis. The type of disease that produces demineralisation is the slow, smouldering, latent condition which may go undiagnosed for years. It must be seriously considered not only in the younger group but in any particularly severe post-menopausal porosis.

One patient (not in this series) was known to have had marked osteoporosis, though without gross collapse, several years before muscular weakness and wasting appeared. Re-examination then revealed nervousness, sweating, tachycardia and goitre. The protein-bound-iodine was 12 milligrams per 100 millilitres. Thyrotoxic myopathy was diagnosed, and all symptoms were eradicated by treatment with radio-iodine.

Ankylosing spondylitis—This is an important cause of back pain and loss of mobility in a young man, and since it is accompanied by considerable spinal porosis it is not surprising that differentiation is sometimes difficult. The presence or absence of radiological evidence of sacro-iliitis and ligamentous calcification will be the final arbiter.

Hypopituitarism and hypogonadism can cause osteoporosis, presumably because of lack of the relevant sex hormone, which is necessary for the proper maintenance of bone matrix. Primary hypogonadism probably needs to be gross to cause any severity of symptoms, and in such cases eunuchoidism is usually obvious. Hypopituitarism may be more difficult to diagnose.

A recent case (not in this series) was that of a woman of twenty-three who developed amenorrhoea three years after " meningitis." Backache was next complained of, and marked osteoporosis was discovered. The skull showed erosion of the sella turcica, and she was found

to have a craniopharyngioma. Presumably the tumour had produced hypopituitarism, with secondary hypogonadism, and this led to osteoporosis.

Osteogenesis imperfecta is believed to be caused by a congenital deficiency in number or activity of osteoblasts, whose job it is to assist in the formation of osteoid; that is in the laying down of bone matrix. Osteogenesis imperfecta is, therefore, a congenital variety of osteoporosis. In distinction from acquired osteoporosis it occurs as a rule at a very early age, even pre-natally; it affects the skull and peripheral bones; it retards growth, and it produces softening of bones with bending as well as fragility. Many patients have no hereditary story, and a few do not have symptoms until adolescence or even early adulthood. These later cases are mild—in some the defect is largely spinal, in others the spine may be normal. When the onset is late and the disorder is mainly vertebral, and in the absence of a hereditary story and blue sclerae, it may be almost impossible to distinguish osteogenesis imperfecta from idiopathic osteoporosis. In fact there may be no tangible difference between them (Dent 1955).

Case of osteogenesis imperfecta mimicking osteoporosis—A white man aged fifty-six was admitted for treatment of barbiturate poisoning. He had fractured many bones easily since childhood, but there was no evidence of fractures or deformities except for bowing and cystic changes in the upper half of the left femur. He had been getting deaf for seven years. The sclerae were dark blue, but there was no laxity of ligaments and his teeth were well formed with good enamel. The spine (Fig. 13) showed moderate porosis, ballooning of disc spaces, variable degrees of partial vertebral compression and secondary anterior osteophytes—a typical picture of osteoporosis.

Comment—The blue sclerae, the history of multiple fractures and the femoral changes pointed away from idiopathic osteoporosis. Lack of much backache and a low urine calcium were also more in favour of the congenital disorder.

FIG. 13

Osteoporotic spine of man aged fifty-six with osteogenesis imperfecta. In appearance it is indistinguishable from that of idiopathic osteoporosis.

ETIOLOGY

Very little is known of the etiology of idiopathic osteoporosis. Various tracks have been followed but most of them ended blindly. The relation to pregnancy has already been considered.

Nutrition—Meulengracht (1939), reporting cases of osteoporosis, did not accurately distinguish the condition from osteomalacia, and believed that they were caused by a deficient diet or disease of the digestive tract. Burrows and Graham (1947), started with this assumption but soon found that it was untenable, and there is no doubt that this is the correct view. In the present series there was no single instance of any digestive disorder such as could cause malabsorption; even achloryhydria was found only once. There was no evidence for deficient diet, except in the single native who, at one time, had suffered from scurvy. In general the patients were not poor, and none was a food faddist.

Occupation and activity—In view of the well known relationship between immobilisation and osteoporosis, it might be wondered whether an active occupation or hobby might protect—in other words, were the sufferers from idiopathic osteoporosis unusually sedentary? This was found not to be the case. The patients include engineers, golfers, athletes, labourers and truck drivers. Four were doctors.

Secondly, had the patients spent long periods in bed or been in some other way immobilised

before the development of the porosis? In no case at all could that be shown. Certainly some had been very inactive, or temporarily bedridden, after an early bout of pain, but in every instance where proof was available, the decalcification was already evident at this time.

Loss of sex hormones—None of the patients in this series was eunuchoid, and, in fact, the osteoporosis of definite and severe hypogonadism is not as pronounced as in the " idiopathic " group. The histories made it quite clear that good libido and sexual activity were usually present while the porosis must have been developing and even after symptoms had appeared. Several fathered children during this period. As noted above, the 17-ketosteroids were not generally depressed. Nevertheless there were a few men in whom loss of libido or sterility for several years was a feature. More definite evidence was sought in some cases by sperm counts or testicular biopsy, but the findings were inconclusive. In Case 2 there was no evidence of testicular atrophy on biopsy, and the patient became the father of a child after the symptoms began. In Case 5 the sperm count was normal six years after the onset of symptoms. In Case 7 testicular biopsy showed " mild hypospermatogenesis with focal atrophy and some tubular sclerosis " (report by Dr Fred Simmons). In Case 9 there were no children in six years of married life; no birth control practised. Sperm count ten million per millilitre, F.S.H. negative at 6½ mouse units; 17-ketosteroids 13 and 14 milligrams in twenty-four hours. In Case 18 there were no children born in ten years of married life (wife tested and found normal). Sperm counts showed defective spermatogenesis. Biopsy showed marked atrophic changes. 17-ketosteroids only 4·4 milligrams. In Case 21 the patient had noticed a recent diminution of libido; youngest child aged four. Biopsy showed bilateral testicular atrophy. 17-ketosteroids were 7·6 milligrams. In Case 27 there had been total loss of libido for seven months. Testicular biopsy was normal; but the patient improved on testosterone.

What relation these genital findings have to porosis is not known, but it seems unlikely that they can be important or causative.

Liver dysfunction—The concept of decalcification due to hepatic disease is ill defined. Theoretically osteoporosis would seem possible in the cirrhosis of severe alcoholism with its protein-starvation effect, or, in the form of osteomalacia, in long-standing obstructive jaundice with steatorrhoea and hypovitaminosis D (Snapper, Seely, Falk and Feder 1954). It must be very rare and certainly played no part in the present series.

Protein loss through the kidney, as albuminuria or amino-aciduria, was never found. Dent (1955) also failed to find excessive loss of amino-acids.

" Pre-osteoporosis " and its relationship to idiopathic hypercalcuria—Calcific renal calculi frequently occur in otherwise normal people with an over-high urinary calcium output (" idiopathic hypercalcuria," Albright and Reifenstein 1948). Others have a similarly high urine calcium without developing stones. In some patients there is an over-absorption of calcium from the gut; others are surely losing calcium from their bones. If such a calcium loss continues for long enough, important and radiologically visible decalcification must result. As already remarked, a clinically porotic patient must have been in negative calcium balance, with high urine loss, for several years before symptoms appear. One may, therefore, talk of a state of " pre-osteoporosis " (as of " pre-diabetes "). If a subject were to be examined in this state, he would be found to exhibit " idiopathic hypercalcuria."

Diabetes—Albright and Reifenstein (1948) and Hernberg (1952) believed that osteoporosis is especially liable to occur in diabetics. There is little evidence for this. A survey being made at present on all diabetics attending our clinic has so far not supported this concept. In the present series only one porotic patient was a diabetic, and the diabetes appeared after the porosis. In eight other patients glucose tolerance curves were made; five were normal and three showed slight prolongation of hyperglycaemia at the two-hour period.

Premature ageing—Some of our patients looked older than their years, but in general the idea that this porosis might be a symptom of premature senility—a sort of late progeria—is not borne out. Certainly those in the earlier stages of the process showed no appearance of

undue ageing. However, our impression is that an unduly high proportion show gross calcification of the larger arteries, as seen radiographically.

Deficiency of bone phosphatase (hypophosphatasia)—Rathbun (1948), Schneider and Corcoran (1950), Sobel *et al.* (1953), and Neuhauser and Currarino (1954) have reported the syndrome of low serum and tissue alkaline phosphatase in infancy giving rise to an odd variety of rickets and defective osteogenesis. Recent studies of phosphatase suggest that it is less important in calcification than in bone matrix formation (Siffert 1951, Pritchard 1952). Consequently it is not unreasonable to presuppose an adult syndrome of acquired deficiency of bone matrix (osteoporosis) caused by lack of phosphatase activity.

Six patients in this series seemed to have a low serum alkaline phosphatase. Over fourteen phosphatase readings were made in one woman (Case 28), and nine were below 4 King-Armstrong units. One man (Case 7) had eight readings taken, of which six were under 1·5 Bodansky units. Tissue was not removed from any patient for assay of its phosphatase content, but it seems quite possible that depressed phosphatase activity may play a part in certain cases of osteoporosis.

Prolonged " alarm reaction " after injury—Albright (1953) suggested the possibility that a prolongation of the nitrogen-loss phase of the alarm reaction after injury might lead to progressive osteoporosis. Thirteen of the patients in this series dated their symptoms from an initial injury which was severe in six, and mild in seven. This idea presupposes, however, that the spine is normal up to the time of the initial injury. But in every case in which radiographs are available at this juncture porosis was already visible. We can therefore produce no evidence in support of this hypothesis.

The idea of a causative injury brings to mind Kümmell's syndrome. Kümmell, in 1895, described cases of injury to the spine followed months later by incapacitating symptoms with kyphosis and gibbus formation due to " disturbance of several vertebral bodies." The conception of this state (originating in pre-radiographic days) has since become nebulous. Most authors (Cardis *et al.* 1928, Rys 1934, King 1935, Hosford 1936, Steel 1951) refer to progressive collapse of a single vertebra, and Kümmell himself, in 1928, admitted that many of his old cases really had tuberculosis. Apparently no writers on this condition comment on the presence of porosis in the spine as a whole, and very few reports have been found in which initial radiographs were described (Steel 1951). It would seem doubtful whether Kümmell's disease as originally formulated really exists, while its modern descendants are so vaguely described that their possible relation to osteoporosis cannot be accurately assessed.

Hereditary factors—There is little evidence to indicate that genetic factors play any part in idiopathic osteoporosis. One man (Case 25) with moderately severe porosis had a brother (Case 26) who had suffered from several bouts of back pain and showed mild decalcification with some compression of the fifth lumbar vertebra. One woman (Case 34) had a second baby who sustained two unexplained fractures of the humerus and one of the femur within the first few weeks of life. Radiographs were said to show porosis, but they were not available for examination; later the baby seemed quite normal.

While, therefore, we can say nothing definite regarding heredity in this state, second cases in families should be looked for.

Conclusion—We cannot really say what causes this disease; maybe different things in different cases. Perhaps there is a congenital weakness in the osteoblasts or in the phosphatase-producing mechanism, and perhaps the porosis is finally precipitated by the intermediation of an accident, pregnancy, mild hypogonadism or other agency.

It may well be that there is no real difference between " idiopathic " osteoporosis and that which occurs in older people. The concept would then be that of a relatively frequent predisposition to osteoporosis, becoming manifest usually in the course of ageing and after loss of sex hormones, but sometimes earlier in life in those more severely predisposed. When younger people are affected by what basically is the same disorder, it is called " idiopathic."

TREATMENT

The treatment of the older osteoporotic was discussed by Moldawer (1955), and the principles apply equally to the sufferer from idiopathic osteoporosis. 1) Immobilisation should be avoided so far as pain allows. The patient must nevertheless be warned not to bend the spine abruptly, and never to strain it while in flexion. 2) Orthopaedic appliances are valuable to allow the patient to get up and about, to give him confidence and a feeling of stability. Braces should be worn only as much as is necessary and the aim must be gradually to discard them. Surgical corsets are usually more comfortable. 3) Dietary supplements of calcium, vitamins and protein are of doubtful value.

Sex hormones—Since most of our patients were sexually active, it was not to be expected that they would respond to sex hormone therapy, in this way differing from the senile and menopausal groups. In fact this is largely true; one man (Case 18), in whom testicular biopsy demonstrated atrophic changes and in whom one might therefore expect testosterone to be helpful, showed no clinical improvement and even a worsening in his calcium balance on this hormone. Balance studies in three cases (reported by Albright and others 1944, 1946, 1948) showed no benefit from male and female sex hormones, even in large doses. On the other hand two patients in this series (Cases 19 and 20) went into strongly positive calcium balance when treated with testosterone, added calcium, and vitamin D together. The patient in Case 27 was most interesting in this respect. Despite lack of evidence of hypogonadism and a normal testicular histology, his urinary calcium output was repeatedly depressed by testosterone, rising again after its cessation (Fig. 14). The effect of testosterone in the female has not been reported.

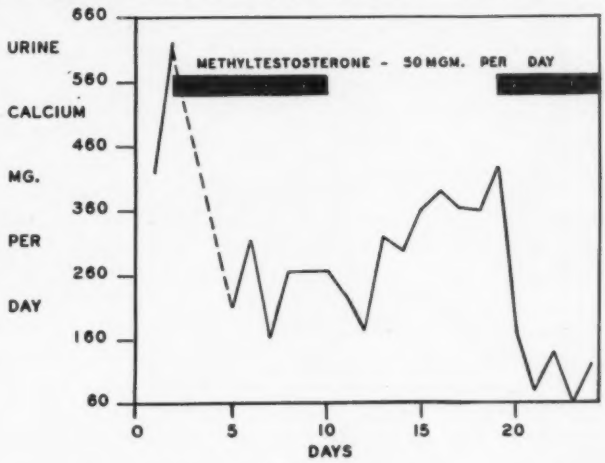

FIG. 14

Effect of methyl testosterone on the urinary calcium in Case 27. Such a satisfying reduction was not expected in view of normal testicular histology. (Note the rise after discontinuation of testosterone and repeated fall when it was restarted.)

Protein—Albright and co-workers (1944, 1946, 1948) showed definite improvement in calcium and nitrogen retention, with concurrent amelioration of symptoms, in three patients treated with repeated intravenous infusions of salt-poor human serum albumin. Dent (1955) claimed improvement in one case with infusions of concentrated plasma, but this treatment failed in others. Infusions of amino-acids have been of no value.

In such a chronic disease this treatment is impracticable except in the acute phases. It might be hoped that a high protein diet would supply the same stimulus to bone matrix formation as serum albumin, but balance studies and the patients' symptoms have shown much less distinct benefit from this.

Vitamin D and calcium—Meulengracht claimed improvement in two patients on vitamin D in large doses and calcium supplements. Two of our patients on this treatment went into positive balance, which was maintained for at least five weeks in one case, and nine in the other.

Conclusions—Despite their doubtful and variable value, I would recommend that a high protein diet (150 grammes a day), containing at least 1 gramme of calcium a day, and vitamin D

(20–50,000 units daily) be tried in all cases. In those with hypercalcuria the urine must be watched to see that its calcium content does not markedly increase, because of the theoretical danger of causing renal stones.

In patients in an " acute phase," intravenous serum albumin or plasma may be tried, but large amounts will be necessary.

PROGNOSIS

None of the patients in this series has died. We have news of twelve of them seven or more years since diagnosis or the onset of symptoms. On the whole the tendency seems to be towards slow symptomatic improvement, with a concomitant lowering of urinary calcium output (Table I).

In general one is impressed with the very small lasting effect that any treatment seems to have had on the long-term cases.

SUMMARY

1. The syndrome of osteoporosis is reviewed and its various causes are mentioned. Osteoporosis in youngish patients without any demonstrable cause is referred to as " idiopathic." The scant literature on this condition is reviewed. Its clinical, radiological, biochemical and histological features are considered.

2. A series of thirty-eight cases is analysed, and illustrative case histories are described. The peculiarities of the disease as it is seen in women are discussed, particularly the relationship to pregnancy and lactation, which appear to act as precipitating factors, rather than being primarily causative.

3. The differential diagnosis is discussed. Osteogenesis imperfecta may not always be easy to distinguish; since it is really a " congenital osteoporosis " this is hardly surprising.

4. The following possible etiological factors are propounded (apart from pregnancy): nutritional, occupational, lack of sex hormone, liver dysfunction, loss of protein, diabetes, premature ageing, hypophosphatasia, " alarm reaction," and inheritance. None of them can be incriminated except in the odd case. The relationship between osteoporosis and idiopathic hypercalcuria is mentioned. The only conclusion regarding etiology is that some people are simply more prone to bone loss than are others, and in these a variety of accentuating factors may render the disorder clinically apparent.

5. The treatment of the condition is unsatisfactory, although occasionally a positive calcium balance may be obtained with sex hormones or intravenous infusion of plasma albumin or whole plasma. The general tendency seems to be towards clinical improvement (biologically " stabilisation " rather than improvement), but some patients become permanently crippled.

Methods—Since the cases in this series were investigated at different centres the biochemical and biological methods were not uniform, and it is felt unnecessary to detail them. All were, however, well known and standardised.

I most sincerely acknowledge my gratitude to Dr Fuller Albright for his inspiration, for permission to study and use cases under his care, and for his valued criticisms of this paper. Dr Albright has given his general blessing to the article, but does not necessarily agree with all the opinions expressed therein. I also wish to thank all the doctors, too numerous to mention individually, who have referred their cases to us. I am indebted also to Professors J. F. Brock and F. Forman for their encouragement and insistence that this paper should be written, to Dr R. Hoffenberg for suggestions and invaluable help with the manuscript, and to Mr B. Todt for the photographs. Acknowledgment is made to the Editor of the *South African Medical Journal* for permission to reproduce some of the Figures.

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THE ILIAC APOPHYSIS AND THE EVOLUTION OF CURVES IN SCOLIOSIS

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In 1948 Risser reported an observation made at the Los Angeles Orthopaedic Hospital that " the completion of ossification excursion of the iliac apophysis has been proven to occur simultaneously with the completion of vertebral growth, and with it the curvature is static . . .". Although this was confirmed by other authors (Steindler 1950, James 1954, Donaldson 1957) no statistical evidence has yet been offered to support the observation, nor have data been presented on the relationship between the excursion of this ossification centre and the evolution of scoliotic curves.

For the surgeon engaged in the management of scoliosis and seeking for a sign that would be decisive for or against operative intervention, the observation made by Risser is of great practical importance.

The aim of this investigation has been to study the radiological characteristics of this ossification centre, its relation to other signs of maturation such as the onset of menstruation and the growth of the apophyses of the vertebral bodies, and its relationship with the progression of the curve in scoliosis.

More than 800 patients with mature curves seen at the Royal National Orthopaedic Hospital in the last ten years were reviewed, and of these, 224 were found suitable for this investigation. In each there was at least one radiograph of the curve available at the time the iliac apophyses had completed their excursion (that is, reached the posterior limit of the iliac crest), and one follow-up film taken not less than a year later.

CURVE PROGRESSION AND THE ILIAC APOPHYSIS

In approximately half of the 224 cases a series of radiographs from the first appearance of the iliac apophyses until after their fusion to the crests was available. Observations on the evolution of curves have been grouped into three: there are the changes in the curve occurring

TABLE I

THE MAXIMUM AND AVERAGE AMOUNT OF INCREASE IN THE CURVATURE OBSERVED DURING THE THREE PERIODS

Periods	Maximum increase	Average increase
1 Between appearance and completion of the apophyses .	38 degrees	13 degrees
2 After completion of their excursion until fusion . .	26 degrees	9 degrees
3 After fusion of the apophyses to the crest . . .	18 degrees (only 1 case over 15 degrees)	8 degrees

between the appearance of the iliac apophyses and the completion of their excursion, the changes after completion until fusion, and the changes after fusion of the apophyses to the iliac crest.

An overall picture of the evolution of curves during the three periods is presented in Figure 1. The term "increased" has been used whenever there was an increase in the angle of curve of more than 5 degrees; "unchanged" curves include cases in which the increase was less than 5 degrees or in which an apparent decrease of a few degrees was observed, a variation often due to measuring errors. In each group the percentage of the total cases in which a deterioration of over 15 degrees had occurred is separately indicated. These figures provide

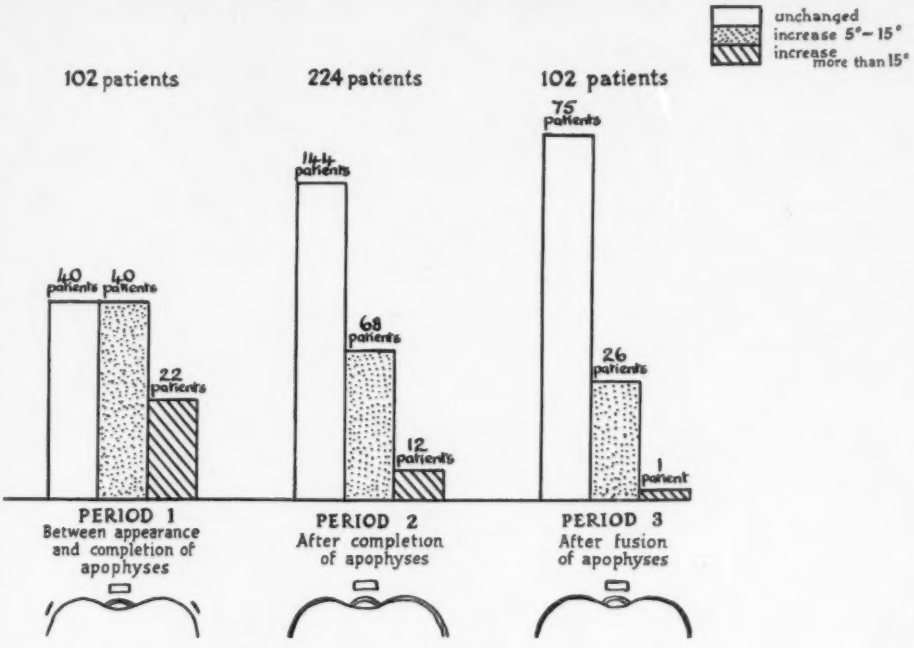

FIG. 1

The evolution of curves during the three periods of growth of the iliac apophyses.

statistical support for the belief that skeletal maturation as indicated by the completion of the iliac apophysis marks the end of significant deterioration in scoliosis.

As can be seen from Table I, even during the first period, before the apophyses had completed their excursion, the increase of the curve was not alarming, averaging 13 degrees. In the two subsequent periods it was naturally less. One should, however, refrain from too much optimism; the increase in each individual period is small, but the total amount of deterioration at the end of the three periods may be significant.

When these cases were analysed from an etiological standpoint it was found that, contrary to the belief of some surgeons, the paralytic curves usually follow the general rule that no significant deterioration occurs after the iliac apophyses have completed their excursion. This is illustrated in Figure 2. Almost half of the cases of paralytic scoliosis were followed long enough (until after fusion of the apophyses to the iliac crest) to demonstrate that deterioration in these curves did not occur at a later stage.

Curves of other etiology than idiopathic and paralytic are not shown in this figure because each group was too small to allow conclusions to be drawn.

THE PRIMARY SITE OF SCOLIOSIS AND THE ILIAC APOPHYSIS

The evolution of curves during the three periods was studied from the point of view of curve pattern. The worst deteriorations during both the period before and after maturation

occurred in the combined (double primary) curves regardless of etiology or of age of onset. In a number of cases, however, only one of the two primary curves had deteriorated whereas the other remained unchanged. During the first period, before completion of the iliac apophyses, the combined curves not only showed the highest percentage of deterioration over 15 degrees,

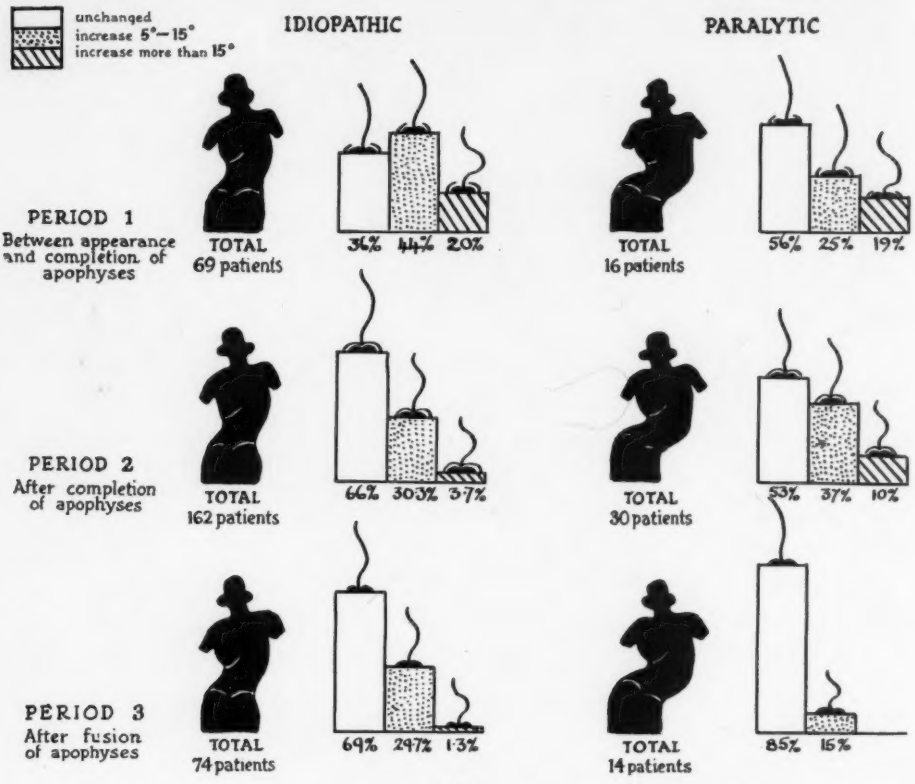

FIG. 2

The similarity of curve evolution in idiopathic and paralytic scoliosis in the three growth periods.

but also the most severe deterioration in individual cases. During the second period the thoraco-lumbar curves also showed a high percentage of deterioration over 15 degrees. Figure 3 summarises the distribution of deterioration over 15 degrees according to the pattern of curve during the periods before and after maturation.

THE OSSIFICATION CENTRE OF THE ILIAC CREST

As expected, considerable variation was found in the time of appearance, completion and fusion of the iliac apophyses. In Table II the rather incomplete data collected from the literature are recorded, together with our own observations.

The radiographs showing the apophyses of all our patients who had paralytic curves were examined separately. It was found that in these patients, too, the apophyses appeared, matured and fused at the times shown in Table II. Only in the female poliomyelitic patients did the apophyses appear and mature a few months later than the average times.

Risser's (1948) observation that a year usually elapses between appearance and completion of the iliac apophysis was confirmed. Our average figures (for boys and girls, Table II) were slightly longer than his.

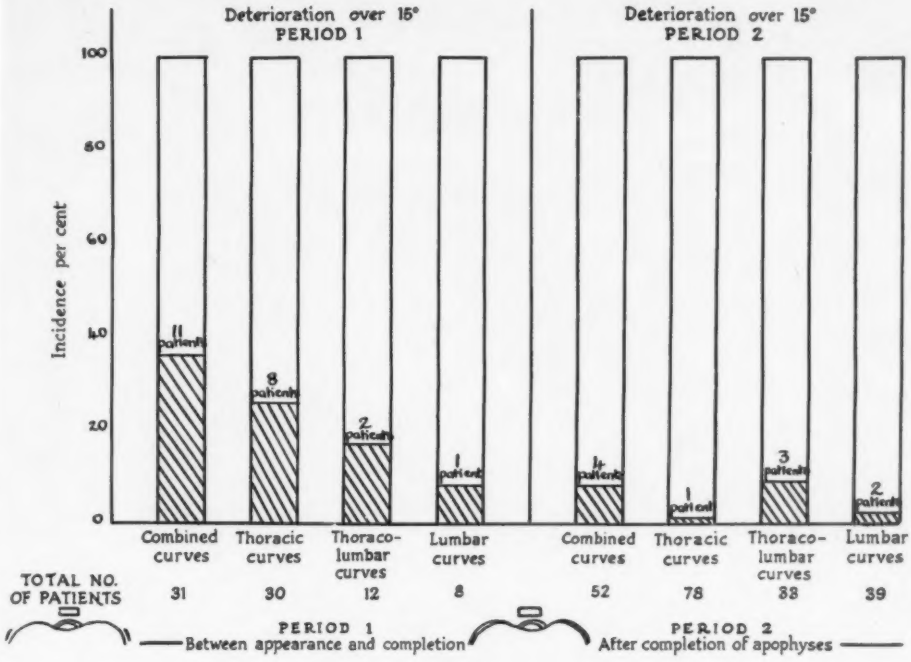

FIG. 3

The incidence of deterioration over 15 degrees in periods 1 and 2. In period 3 there was only one such case among 102 patients.

In our series the longest period that elapsed between appearance and completion of an apophysis was three and a half years for both boys and girls. The shortest was seven months for both sexes. The apophysis fused to the iliac crests within an average interval of two years after the completion of its excursion in girls, and a little more quickly in boys. The most

TABLE II

THE TIME OF APPEARANCE, COMPLETION AND FUSION OF THE ILIAC APOPHYSES

Author	Average and extremes	Age of appearance (*years*)		Age of completion of excursion (*years*)		Age of fusion to the crest (*years*)	
		Boys	Girls	Boys	Girls	Boys	Girls
Waldeyer (quoted by Schinz)		15–16	15–16	—	—	24–25	24–25
Flecker	Average	16	14	—	—	21	21
	Earliest	13½	12	—	—	—	—
Köhler		12–15	12–15	—	—	21–25	21–25
Zaoussis and James	Average	15½	14¼	16½	15½	18¼	17½
	Earliest	12¾	11	13	12½	15¾	13½
	Latest	17½	18	19¾	19¾	21¾	23½

rapid fusion of an apophysis to the crest was observed in a boy five months after the apophysis had reached the posterior limit; the shortest period observed in girls was six months. The longest time an apophysis took to fuse in this series was six years in a girl, whereas the longest for boys was two and three-quarter years.

Again there was no difference noted when the rate of growth of the apophysis in patients suffering from poliomyelitis was investigated.

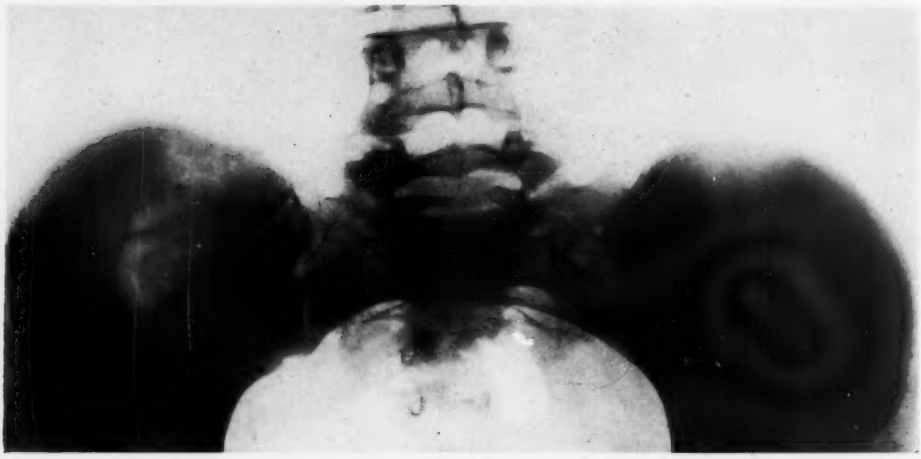

FIG. 4

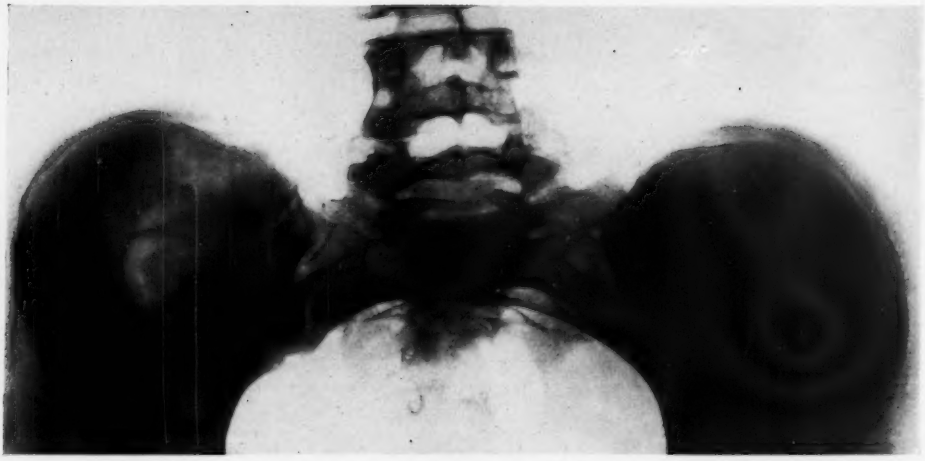

FIG. 5

Figure 4—A view of the iliac apophyses using an ordinary viewing box. Figure 5—The same radiograph in a bright light. The " absent " apophyses have almost completed their excursion.

DEVELOPMENT OF THE ILIAC APOPHYSES

For the precise assessment of the state of the apophysis the radiograph must be examined in a bright light (Figs. 4 and 5).

In most cases the apophysis makes its appearance at the anterior part of the iliac crest. It then grows backwards along and parallel to the iliac crest (Figs. 6 to 8). Although anatomically

FIG. 6

FIG. 7

FIG. 8

Figure 6—The apophyses are just visible. Figure 7—The apophysis has "dipped down" on one side. Figure 8—The apophyses are fusing.

the apophysis reaches the posterior superior iliac spine (Fig. 9) this is seldom seen in the ordinary antero-posterior view of the spine which includes the iliac crests. The explanation is that the posterior superior spine lies much lower in relation to the iliac crest than is usually thought, and cannot be well shown. For this reason it was decided to define the completion of the apophysis as the time at which it dips down to make contact with the iliac crest at its posterior limit, as described also by Risser (Fig. 7).

Of 224 cases there were only six (2·6 per cent) in which the apophysis could be seen on one side only. But even in these cases a small ossification centre would almost invariably appear in the naked crest at about the time the opposite apophysis had completed its excursion or fused to the crest.

Whereas a unilateral apophysis is virtually non-existent, asymmetry in their development along the crest was quite frequent. Figure 10 illustrates a typical example of the difference in progress on each side of the pelvis.

In seventy-four cases definite asymmetry in the development of the iliac apophyses was observed. Since the number of cases available for observation of the full evolution of the apophyses was only 102 (Fig. 1) it must be concluded that the incidence of asymmetrical development is high. However, in only twenty-eight out of the seventy-four cases did this asymmetry persist until the time of completion or of fusion of the apophyses to the crest. In forty cases the apophyses had become symmetrical at about the time they had reached the posterior limit of the crest. In six cases the development became asymmetrical only at its last stage, shortly before completion.

In about 15 per cent of the cases in which asymmetrical growth was observed it was noticed that after the one apophysis had made contact with the crest the curvature increased significantly (over 15 degrees). Even in the remaining cases there was usually a slight increase of the curvature while one apophysis was still developing. It seems, therefore, that prognosis as regards deterioration of the curve should depend on the state of maturation of the less advanced apophysis.

Figure 11 shows another interesting feature in the development of the iliac apophyses. An apophysis with a short excursion is illustrated; and sometimes this seems to fuse with the ilium as it grows. There were only eleven cases (10 per cent) in this series in which one or both apophyses showed a true short excursion. The incidence is identical with the figure given by Risser (1948). A much higher number (twenty-five cases or 24 per cent) showed a slow development with almost simultaneous fusion to the crest. In some the apophysis at an early stage was growing so near the anterior part of the crest that it could be overlooked, and recorded as absent. Suddenly a year and a half or two years later, even though there was no evidence of it in previous serial films, the " absent " apophysis would be discovered fully grown to the posterior limit and fused!

The incidence of a posterior ossificaton centre was much higher in our series than the 1 per cent reported by Risser (1948). In ninety cases (40 per cent) there was radiological evidence of a small or large centre clearly separate from the anterior centre (Fig. 12). The incidence was estimated on the total of our material (224 cases), as the presence of a posterior ossification centre can be identified even at the time of completion of the apophysis. Usually the anterior centre appeared first, but in a few cases the posterior ossification centre was at first the only evidence of the presence of an iliac apophysis. Often the posterior ossification centre will dip down to make contact with the posterior limit of the iliac crest while the main part of the apophysis is still developing along the crest. In view of the high incidence of a posterior ossification centre it was thought worth while to attempt to assess the progress of the curvature from the time of the appearance of this centre in the hope that this might be of prognostic value. The period of study extended from the date when this centre was clearly visible, regardless of whether or not it was in contact with the crest, to the time when the apophysis had either completed its excursion or fused to the crest—in other words up to the

FIG. 9
Shows the apophysis in its complete form reaching the posterior superior iliac spine.

FIG. 10
A typical example of asymmetrical development of iliac apophyses.

FIG. 11
An apophysis with a short excursion.

very end of the apophysial growth. It was found that the average increase of a curvature during this period was no more than 6 degrees, the greatest increase observed being 28 degrees. Only 13 per cent of these cases showed a deterioration of more than 15 degrees, which makes this sign less reliable than the completion of the apophysis (6 per cent of cases or 12 patients increased more than 15 degrees (Fig. 1)) but still a valuable observation.

The most unusual anomaly in the development of an apophysis was observed in one case in which a true double excursion could be seen in serial films (Fig. 13).

FIG. 12
A posterior ossification centre of the iliac crest, seen on one side.

FIG. 13
An apophysis with a double excursion.

CORRELATION OF THE ILIAC APOPHYSIS WITH THE RING APOPHYSIS OF THE VERTEBRAL BODY

In only a small number of cases was the series of radiographs comprehensive enough to permit a comparative study of the two groups of apophyses. Nevertheless it was possible in 156 cases to obtain at least one lateral view of the spine in which the vertebral apophyses were shown at some stage of their growth, and to compare it with the available films of the iliac apophyses. The views obtained were usually of the thoracic region, and in a number of cases

there was in addition a view of the lumbar region. The lumbar vertebral apophyses were almost invariably slightly ahead of the thoracic in maturation.

In Figure 14 the combinations of stages of the two types of apophyses as recorded in our series are presented in order of frequency. In only five cases was a truly synchronous evolution of the vertebral ring apophyses and the iliac apophyses seen. In the remaining

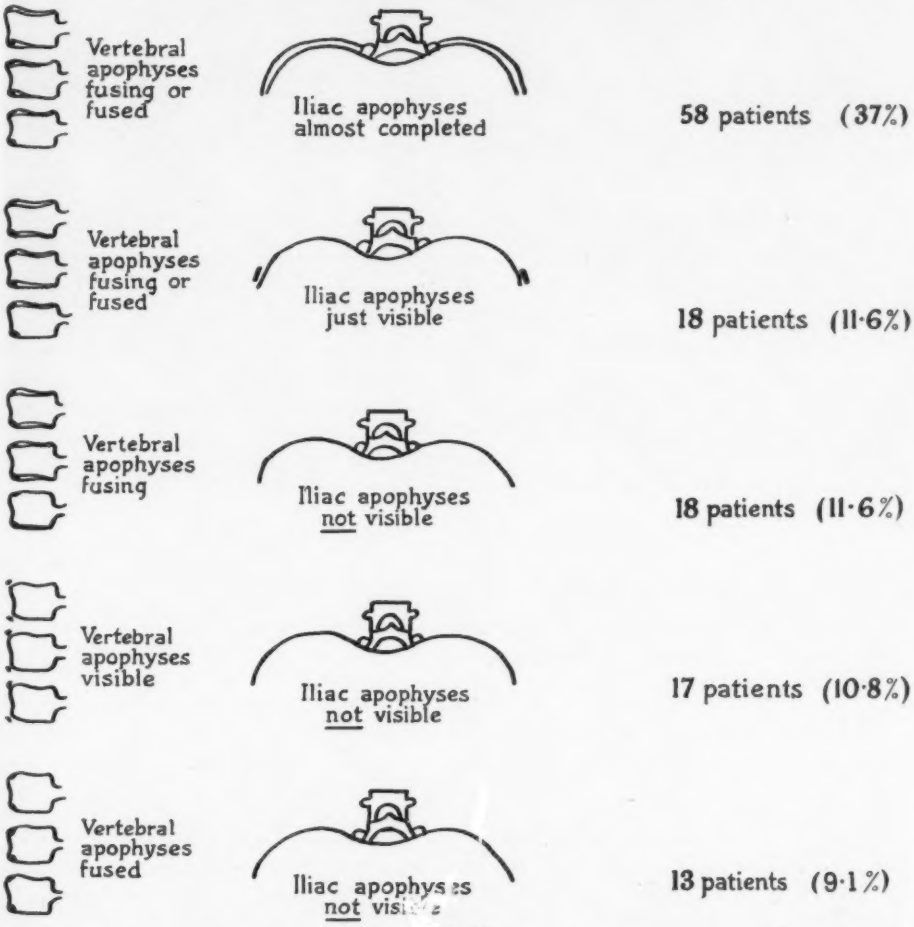

FIG. 14

The growth correlation between vertebral and iliac apophyses. Not included in the diagram were five cases of synchronous development and thirty-seven in which the first radiographs already showed fusion of both.

thirty-seven cases the radiograph available for comparison showed that fusion of both vertebral and ring apophyses had already occurred, but films to demonstrate their earlier development were not available.

The vertebral apophyses appeared at an average age of a little over twelve years, the earliest appearance being observed at the age of nine and a half years and the latest at fifteen and a half years. It was difficult to assess the age at which the apophyses began fusing to the vertebral bodies, because in one and the same film one could, for example, observe the higher thoracic apophyses still clearly separated from the bodies whereas at a lower level the apophyses

had begun to fuse. Probably the average age at which fusion began was a little over thirteen years. It was also our impression that, once it had begun, fusion of the ring apophyses with the bodies was such a slow process that the more rapidly growing iliac apophyses ended by completing their fusion almost simultaneously with the vertebral apophyses.

TABLE III

EVOLUTION OF CURVATURES IN RELATION TO THE GROWTH OF THE VERTEBRAL APOPHYSES

Period	Maximum increase observed	Average increase	Percentage deterioration over 15 degrees
Period 1—Between appearance of vertebral apophyses and skeletal maturation (as indicated by fusion of the iliac apophyses): 31 curves studied	54 degrees	26 degrees	74 per cent
Period 2—Between the stage of commencing fusion with the vertebral bodies and skeletal maturation (as indicated by fusion of the iliac apophyses): 40 curves studied	54 degrees	12 degrees	27 per cent

Table III summarises the deteriorations of curvature observed in relation to the growth of the vertebral apophyses. It was thought that two periods were worth study: 1) the period between the appearance of the vertebral apophyses and the termination of skeletal growth as indicated by the completion or fusion of the iliac apophyses; and 2) the period between the time of commencing fusion of the vertebral apophyses and the termination of the iliac apophyses. The group commencing fusion was selected for the study because there is no stage in the evolution of vertebral apophyses comparable to the stage of completion of the iliac apophyses' excursion.

These figures prove that significant deterioration of the scoliotic curve may occur after the appearance of the vertebral ring apophyses or even after they have begun to fuse. It is therefore not so reliable a prognostic feature as the iliac apophysis. Technically it also involves additional lateral radiographs of the spine, whereas the conventional antero-posterior view of the scoliotic curve usually includes the iliac crests.

RELATIONSHIP OF THE ILIAC APOPHYSES TO THE ONSET OF MENSES

The age of menarche was recorded in practically all of our female patients. The average age was thirteen and a half years, which is a normal figure for this climate and race. This

TABLE IV

EVOLUTION OF CURVATURE AFTER THE ONSET OF MENSES

Changes	From the onset of menses to skeletal maturation	Between appearance and completion of iliac apophyses	After completion of iliac apophyses
Maximum increase	57 degrees	38 degrees	26 degrees
Average increase	15 degrees	13 degrees	9 degrees
Percentage of deteriorations over 15 degrees	41 per cent	22 per cent	6 per cent

means that menarche was on an average nine months ahead of the appearance of the iliac apophyses and two years ahead of the average time of completion of the apophysial excursion. Unfortunately there were only forty-one cases in the series in which radiographs of the curve were available dating back to the onset of menses. Hence the study of the menstrual history in relation to prognosis of the scoliosis was rather limited in the present investigation.

In Table IV the worsening of the curve during the period between the onset of menstruation and the termination of skeletal growth (in the sense described previously) are presented. To facilitate comparison, the figures of periods 1 and 2 in the evolution of the iliac apophyses are included in this table.

The high proportion of cases with a significant deterioration after the onset of menstruation tends to support our impression that menarche is not so accurate a prognostic sign as the completion of the iliac apophyses.

SKELETAL AGE AND ILIAC APOPHYSES

The skeletal age was determined from Greulich and Pyle's *Radiographic Atlas of Skeletal Development of the Hand and Wrist* in thirty of 224 cases, studied usually because delay in skeletal maturation was suspected.

In nineteen cases the skeletal age was found to be behind the chronological. The discrepancy ranged from several months to as long as three and a half years. (In seven cases chronological and skeletal ages coincided, and in four cases skeletal age was slightly ahead of the chronological.)

It is of interest that in more than half of the nineteen cases in which skeletal growth was retarded the iliac apophyses appeared at or slightly before the normal average age, and furthermore that in fourteen cases the excursion of the iliac apophyses along the crest was completed in the normal average time. This may indicate that even in cases with slow skeletal maturation, as determined at the wrist, the iliac apophysis remains unaffected in its growth.

SUMMARY

1. The belief that the cessation of spinal growth and curve progression coincides with the completion of growth in the iliac apophyses has been confirmed in a review of material from 224 cases. This applies also to paralytic curves.
2. In a high number of cases this ossification centre showed an asymmetrical development on the two sides of the pelvis. The appearance of a separate posterior centre of ossification is also common, and probably represents an advanced stage in the growth of the iliac apophysis.
3. Menarche and the growth of the apophyses of the vertebral bodies almost always occurred in advance of the iliac apophyses. They should be regarded as early signs of maturation, not reliable in the prognosis of curve progression.
4. The growth of the iliac apophysis appeared to be unaffected by poliomyelitis.

The authors would like to express their thanks to Mrs Glen Haig and the Records Department of the Royal National Orthopaedic Hospital for their kind help in collecting the material, and to Mr R. J. Whitley for the reproduction of the radiographs.

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OBJECTIVE METHODS FOR DETERMINING THE FUNCTIONAL VALUE OF SENSIBILITY IN THE HAND

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It has been borne home to me with the passage of the years how little the results of the customary tests of sensibility in an injured hand correspond with the actual ability of the patient to use his hand. When one wishes to study cutaneous sensibility from the point of view of function, little is gained by analysing the different modalities. What is desired is the answer to these questions: 1) which of its many tasks can the hand do with the sensibility remaining? and 2) is the lost sensibility returning?

FIG. 1

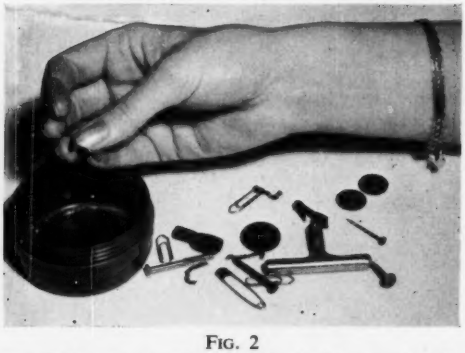

FIG. 2

The picking-up test. Figure 1—Hand with normal tactile gnosis. Figure 2—If the sensibility in the median nerve region is impaired the subject grasps an object with the thumb, together with the ring and little fingers. If the median nerve lacks tactile gnosis it cannot localise or identify objects. This area is "blind."

In Sweden persons receiving workmen's compensation generally return for re-examination every year or so for a long time to have their compensation adjusted, and I have taken this opportunity of making a systematic analysis of the value of the customary tests for judging sensibility.

Partly for this purpose, I have worked out a new functional test for examining the sensibility in an injured hand—that is, a hand with enough motor function to make its sensibility of use. I have called it "the picking-up test," and it is illustrated in Figures 1 and 2. The subject is asked to pick up a number of small objects on a table and to put them as quickly as he can into a small box, first with one hand and then with the other. After he has done this a few times he is asked to do the same thing blindfolded. It is then studied how rapidly and efficiently he picks up the objects; comparison is made between his right and left hands, and likewise between his performance when he is blindfolded and when he is not. The test with blindfolding can be made harder by asking him to identify the objects as he picks them up. If his hand possesses normal sensibility, it can "see" even when the subject has his eyes closed. If sensibility in the median nerve region is impaired, the subject grasps the objects with the thumb and the ring and little fingers (Fig. 2) instead of with thumb and forefinger as he normally would.

I compared the results of this test and of others with the results of currently used tests for sensibility in a number of cases with good motor function but with reduced sensibility in the median nerve area.* The result was astonishing. Most of the current methods proved

* A study to be published elsewhere.

of no value for judging the cutaneous sensibility from the point of view of function. The tests with cotton wool and pins for touch and pain, and the ordinary methods for testing the

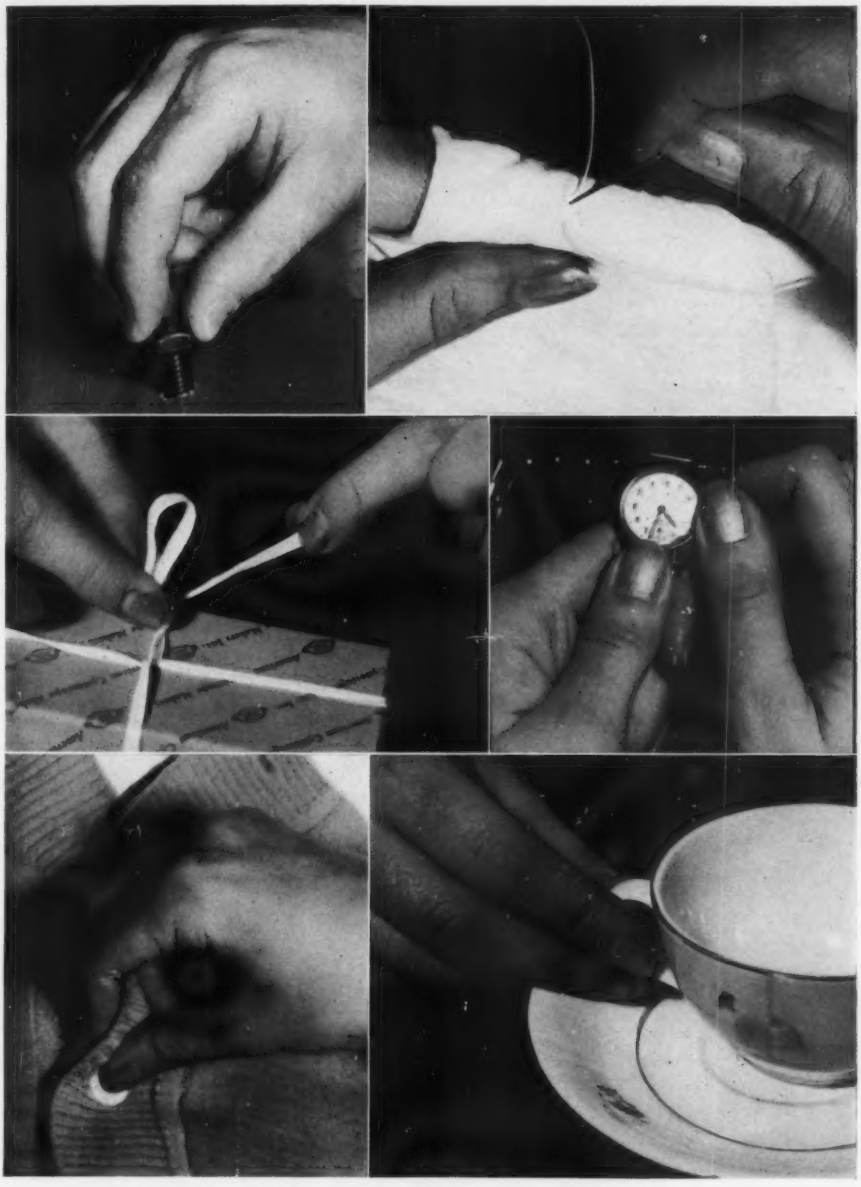

FIG. 3
Precision-sensory grip. Tactile gnosis is necessary for these manoeuvres.

sensations of warmth and cold and deep pressure, were clearly not satisfactory for estimating the functional loss. On the contrary, these methods were apt to be misleading, for one is

prone to believe that a hand that shows a normal or low threshold for touch and pain must also have sensibility of comparable functional value. The fact is, however, that even when it does, and even when it has normal motor function, it may still be quite unable to do the everyday tasks requiring precision—to perform what I have called a precision-sensory grip (Figs. 3 and 4)—and so be greatly disabled.

The only two of the earlier methods that gave useful information about the functional value were Weber's two-point discrimination test and Seddon's coin test. These methods, however, have the disadvantage that they are subjective, and the results depend largely upon how much the examiner and patient are able to concentrate. They are apt to give a false impression, especially if the patient lets himself be influenced by hopes of remuneration. Moreover, they are of no use when the patient is very young, feeble-minded or unable to co-operate for some other reason.

It seemed essential, therefore, to find an objective method of testing sensibility. I observed that an area of skin in the hand which lacks tactile gnosis* also lacks the ability to perspire. Bunnell (1944) wrote: " The skin in an anaesthetic area is so characteristic that one can, by feeling with the finger, determine the area almost as accurately as by testing with an applicator. The finger glides over it smoothly with a satiny feel; it does not jump along as it does on normal skin, in which the sweat glands are working. Atrophic skin appears smooth." Since an applicator cannot be used to differentiate between good and defective tactile gnosis, it is in fact far less exact than the palpating finger.

In my series it was possible to see changes in an area of defective sensibility with the naked eye. Thus it could be seen that the pulp was atrophied, the ridges were more or less eradicated and the colour was changed.

It should be noted that these changes were seen in every case in which tactile gnosis was lacking, provided there was not too much fibrous induration or scar formation. Richards (1954) did not always find trophic lesions of this kind in cases of war injuries in which the sensory innervation was lost, but this was probably because in his cases the picture was frequently confused by scars and other complications.

It is known that, when a sensory peripheral nerve is severed, the skin in the involved region becomes dry only a few minutes afterwards (Richards 1954). The sweat glands stop functioning entirely. I have often observed how rapidly this phenomenon sets in. On the other hand, an area that has sudomotor function and feels moist also has tactile gnosis, apart from the exception to be described later. My experience that the loss of tactile gnosis could be established by palpation formed the background of my attempts to find a method to register the sensibility in the hand objectively, without the co-operation of the patient. In order to be able to assess the results objectively a method of registering the changes had to be found, as palpation is a subjective method of examination.

My first attempt to register the changes in skin areas with defective sensibility was based on fingerprints. I found that it was often possible to map out atrophied areas in the pulp and defects in sensibility with the ordinary fingerprints used for identification. Thus atrophy of the pulp caused the lines of the fingerprint to come closer together, crowding a greater number of lines into a given space. The lines also tended to disintegrate into points. New wrinkles crossing the lines appeared. These changes have apparently never been described in criminological literature, and they proved to be of interest in those circles. However, they do not become manifest until some months after the nerve injury. There proved to be better

* The terms used for this function are both ambiguous and inexact. Here is not a question of the known modalities of sensibility. It is not certain that the sum of these modalities is equal to the total sensibility. The term " stereognosis " is often used in the literature on hand surgery, but in neurological literature it is used for a central function. It is not advisable, therefore, to use it for a constellation of peripheral sensory factors. Moreover stereognosis is not a wide enough concept. Here it is a question of the complex sensibility that gives the grip " sight." For these reasons I have chosen the term " tactile gnosis " (Broman 1945). Tactile gnosis should not be confused with tactile sensibility, a term used for the simple appreciation of touch.

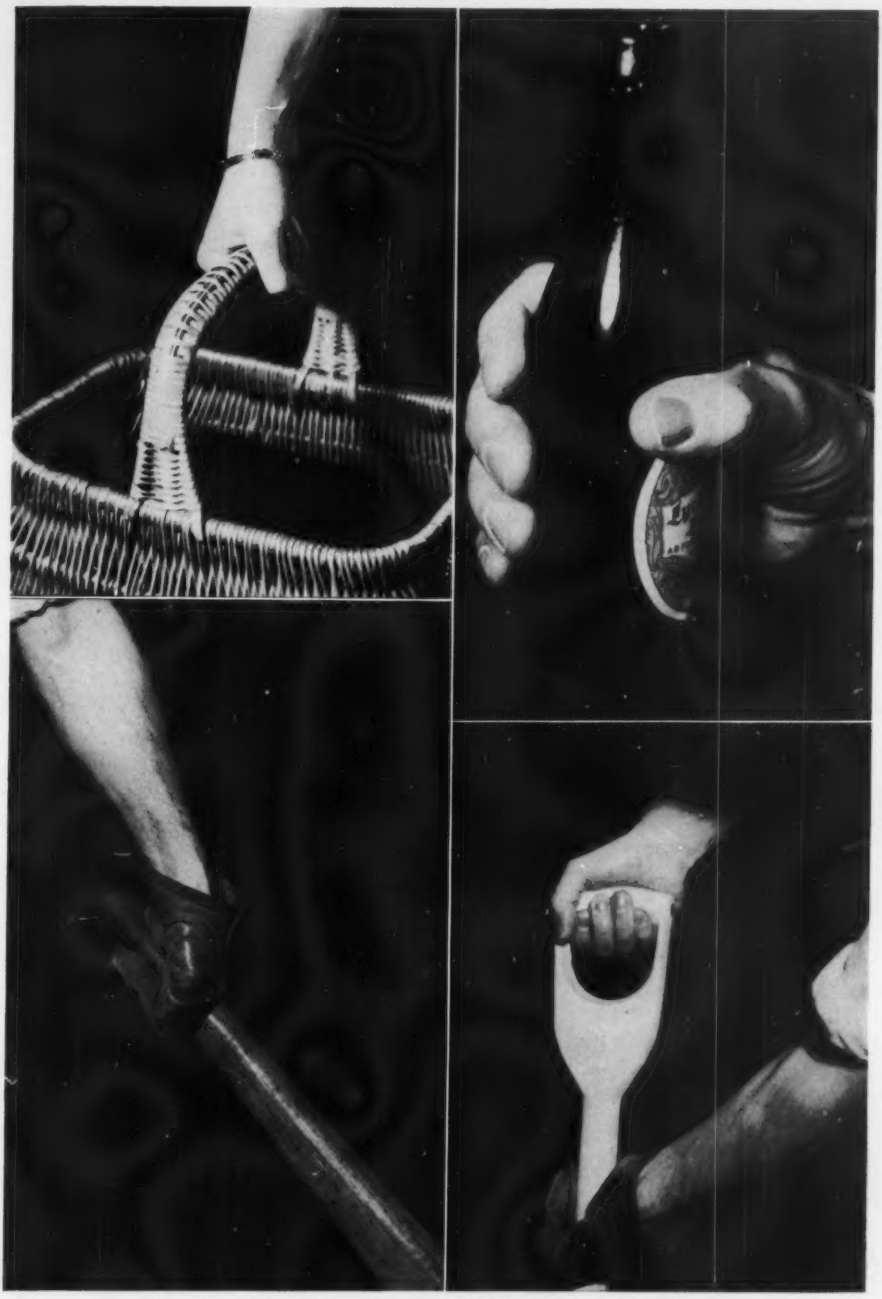

FIG. 4

A gross grip may be useful with a lower degree of sensibility than needed for the precision-sensory grip, but cannot be executed with normal speed and skill.

ways of registering denervation in the skin objectively, based on the changes in sudomotor function between areas with normal and damaged sensibility.

It is by no means a new procedure to register cutaneous sensory loss by demonstrating changes in the sudomotor activity. Three different lines have been followed.

1. Dye tests have been used for many years to study the secretion of sweat. In most of them the skin is first coated with the dye and the patient is made to sweat either with the aid of heat or with different drugs. When sweating sets in, the areas with normal sudomotor function become coloured while the others remain unchanged. Minor (1928) described a method in which the skin was painted with a suspension of iodine and starch in oil. His test has been widely used, but not for testing the sensibility in the fingers and hand. Dorscheid (1954) tried to show hyperhidrotic variations in hand sweating in psychiatric patients with this method. In 1940 Guttmann described a new method using quinizarin. This method has been widely used for testing the extent of sensory loss in large areas of skin. However, it has the disadvantage that the dye is apt to spread about over the patient and the room in which the tests are made. It also causes the patient great discomfort as he has to be kept for some time in a hot chamber. Seddon, Medawar and Smith (1943) used Guttmann's method to follow the degree of " academic recovery " after nerve suture—that is, the restoration of sensibility as shown by neurological methods. To their disappointment, however, they found that the method was " erratic, and therefore quite useless for the estimation of rates of regeneration." Klar (1955) and others have used essentially the same method for diagnostic purposes but not in the hand.

2. It has been known for some time that denervated dry skin and normally moist skin differ distinctly in electrical resistance. The study of this phenomenon, called dermometry, was again popularised by Richter and Katz in 1943. As a rule, they observed a sharp borderline between normal and abnormal regions. Their method has the advantage that it can be used for smaller surfaces than the aforementioned dye tests because there is not the same risk of contamination between perspiring and dry areas. It can also be used when it is difficult to get the patient to co-operate—for instance, for children and in the region of an open wound. Seddon (1954) pointed out that the electrical method of examination is less sensitive than Guttmann's dye test because it is unable to show the pin-point perspiration that may be a sign of an incomplete lesion or incipient recovery.

3. Kahn (1951) showed that one could differentiate between denervated and normal regions by examining them under the microscope after the patient had been made to sweat profusely.

None of these methods has been adopted for practical study of the sensibility in the hand. The reasons are several. The dye tests, especially, cause the patient considerable discomfort, and before the test the skin must be treated with a substance which may affect the sweating to be examined. These methods do not draw a distinct borderline between regions with different sweating reactions, which is essential for the small areas involved in the hand.

Hitherto, and this is an important point, it has been thought that the neurological methods were able to give a differentiated picture of the sensibility in the hand, and that testing of the sudomotor function could only give a simple " yes " or " no." For this reason tests based on the sudomotor function have been considered inferior for determining sensory function. No attempt at all seems to have been made to determine what qualities of sensibility might correspond to the sudomotor function. My aforementioned study, however, showed that whenever an area of the hand showed normal sudomotor function it also possessed normal tactile gnosis. This opened up a new perspective, and I began a search for a method of registering changes in sudomotor function that was more sensitive and exact than the previous ones, but at the same time simple to perform. Two paths led to two different methods. These methods I have called the ninhydrin and iodine-starch printing tests.

PRINTING TESTS

Anatomical background—The secretion of sweat is regulated by the sympathetic nervous system. The fibres of this system enter the brachial plexus in the cervical region far proximally in the form of post-ganglionic fibres (Fig. 5), and then follow the sensory pathways to the periphery. When a peripheral nerve is injured the sweat glands in the skin of the region supplied by this nerve lose their innervation. Only the proximal parts of the brachial plexus, proximal to the level at which the sympathetic fibres enter, can be injured without damaging the corresponding sudomotor function in the peripheral region. All the techniques and conclusions now to be described, therefore, apply to injuries to the peripheral sensory nerves distal to this point in the arm. The methods are of no help when the most proximal part of the plexus or the central nervous system is injured.

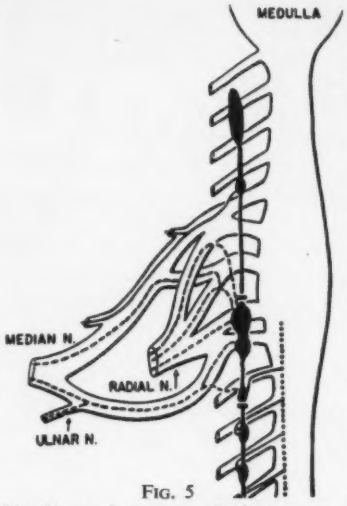

FIG. 5

The fibres of the sympathetic nervous system (interrupted lines) enter the brachial plexus high in the cervical region and follow the sensory pathways out to the periphery. (After White, Okelberry and Whitelaw 1936.)

This exception is of no great importance from a practical point of view, for the great majority of the injuries in question affect the peripheral nerves at a more distal level.

Principles and technique—With both the ninhydrin and odine-starch printing methods, the orifices of the functioning sweat glands are shown in prints of the pulps of the fingers on a strip of paper. The site of a nerve lesion is usually known or is indicated by the presence of a tender neuroma or a scar in the skin. Consequently, the cutaneous sensibility of the skin between the pulp and the nerve lesion can be judged from the prints (Fig. 6).

Ninhydrin method—Hier, Cornbleet and Bergeim (1946) found ten different amino acids in sweat. They calculated that the average cubic centimetre of sweat contained 0·44 milligrams of amino acids. Because of the way they collected their material, however, some of it may have come from the epithelium rather than directly from the glands. Pankow (1953) found the following amino acids in human sweat: asparaginic acid, glutaminic acid, serine, thionin, alanine, valine and methionine. Král and Zenísek (1955) demonstrated that the amino acids excreted in sweat increased with heavy labour.

Ninhydrin stains amino acids and lower peptides with great sensitivity. Odén recognised the value of this for criminological purposes. He and Hofsten (1954) discovered that a moist finger pressed against paper left a print which could be dyed with a ninhydrin solution many years later. They sprayed the paper with a 0·2 per cent dilution in acetone, acidified by glacial acetic acid, dried it in air and then warmed it for a few minutes at about 120 degrees centigrade. They then fixed the print by treating the paper with a solution of copper nitrate.

Odén and Hofsten worked out their method for bringing out old fingerprints for identification. I found that the same principle could be used for testing sensibility in the hand.

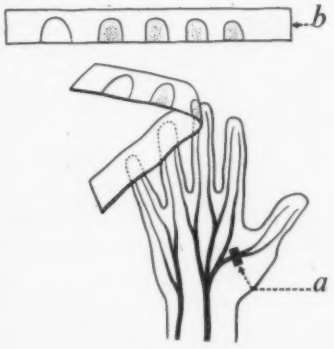

FIG. 6

The printing tests register the functioning glands as dots, and consequently the state of tactile gnosis in the finger pulp. (*a*, nerve lesion; *b*, prints showing loss of tactile gnosis in the pulp of the thumb but normal sensibility in the fingers.)

What is wanted for testing sensibility is not a complete fingerprint, but a punctiform representation of the glandular orifices of the finger pulps. The ninhydrin printing test I worked out aims to show as many of the functioning glands as possible in printed dots. The same is true of the iodine-starch printing method.

Water proved to bring out substances stainable with ninhydrin from the surface of the skin, even when the sudomotor function was completely absent; so the paper must not be moist, and there must not be any extraneous moisture on the hands. It is necessary, of course, to use paper which does not contain substances stainable with ninhydrin in the glue or elsewhere. The method is so sensitive that the examiner must be careful not to touch the strips with his hands, or else unwanted prints will appear after a few days, even if the paper is not heated.

Iodine-starch printing method—Randall (1953) modified Minor's test by having the starch incorporated in a paper and pressing the skin against this paper. Dole and Thaysen (1953) improved the method by having both the starch and iodine put into the paper. When the skin was pressed against this paper the water in the sweat caused blue spots on the paper, the size of which corresponded to the amount of water given off by the skin. They claimed that they could get a print from every functioning gland with their method. They did not use it for studying sensibility, but with minor modification it proved to be valuable for my studies.

Details in the performance of the tests—It must be emphasised that exact and clean work and training are always necessary to get reliable prints. The prints are obtained by pressing the pulps of the fingers, one at a time, steadily against a strip of paper measuring about fifteen centimetres by three. The outline of the finger tip is traced with pencil (containing no dye) on the paper. Sometimes it is useful to roll the tip of the fingers slowly and steadily from side to side so as to get its lateral surfaces on the print, especially in the case of the thumb where the border zones innervated by the radial nerve are of great practical importance.

The secretion of sweat in the pulps must not be too intense. Impressions like that shown in Figure 7 are the ones desired. Skin too moist leaves blurred impressions, as in Figure 8, and makes it difficult or impossible to draw an exact line between areas of normal and reduced sensibility. The secretion from strongly sweating regions is apt to contaminate the dry areas. In such a case the skin should be washed with soap and water, ether or alcohol, and then well dried.

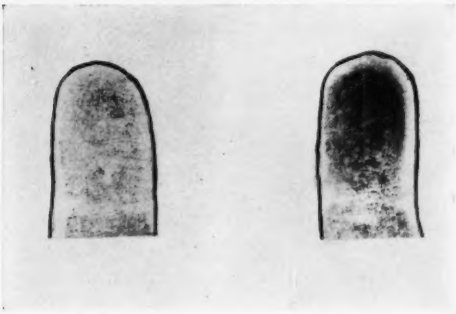

FIG. 7 FIG. 8

Figure 7 shows the sweat gland openings as dots. This is the type of print desired. Figure 8 is from too moist skin, and is too clumsy to permit exact evaluation.

The secretion must not be too weak, otherwise there will be too few dots, or none at all. When the patient is cold or has been given medication, or during anaesthesia, he may perspire so little that not even a normal area leaves a print. Then the secretion must be stimulated. Salicylic acid, warm tea and exercise are helpful. Pilocarpine has been found to be of no value. For special studies intense stimulation can be induced by a subcutaneous local injection of mecholyl (beta-acetyl-methyl-choline-hydrochloric acid).

As a rule, however, good prints are obtained in the ordinary room temperature in Sweden (about 18 degrees centigrade—64·4 degrees Fahrenheit) without previous preparation of the patient.

If there is a contracture or some other deformity in the finger, it may be difficult to take the print in the way just described. Then a piece of paper the size of a postage stamp can be pressed against the finger pulp, either with a piece of rubber sponge, or in a special metal

holder (Fig. 9) to which an elastic band has been attached so as to give a cushioned printing surface. It takes practice to get good prints in cases of this kind.

Prints can also be obtained from parts of the hand other than the pulps of the fingers, but as a rule nothing is gained from taking a print of the whole hand.

For the ninhydrin printing test several ordinary kinds of white writing paper proved useful. The paper recommended later for the iodine-starch method stained in a disturbing way. Porous paper is not good. The best way to develop the prints is to dip them in a 1 per cent ninhydrin solution in acetone, which will keep for several months. About ten millilitres are mixed with a few drops of glacial acetic acid in a cuvette. The acidulated solution will only keep for one or two weeks. The paper strips dry quickly, and after drying they are warmed in air at 100–120 degrees centigrade for five or ten minutes. Thereupon the dots become visible, but it is advisable to wait a few days before fixing the prints, because they often grow more distinct. They are fixed by dipping them in the solution described by Odén—a 1 per cent solution of copper nitrate in a 5 : 95 mixture of water and methyl alcohol or acetone, acidified by a few drops of concentrated nitric acid per hundred millilitres.

FIG. 9
Metal holder with a rubber band for taking prints from fingers with a flexion contracture.

For the iodine-starch printing test, it is important to get suitable paper. Dole and Thaysen (1953) used " 80 lb. substance, white richfold coated (Rex Stationery Co., New York City)," and it is the only type with which I have been able to get good results. The strips are put over a filter paper uppermost in a petri dish on the bottom of which are placed a number of iodine crystals. The dish is covered with a lid and heated at about 60 degrees centigrade for a few minutes. The iodine becomes sublimated and stains the strips yellow. It makes no difference if the staining is slightly uneven. The paper quickly loses the iodine; so the strips should be used soon after they are prepared. Distinct prints can be obtained, which are easy to study when they are moderately enlarged.

Comparison between iodine-starch and ninhydrin printing methods—The iodine-starch method has the advantage that the print can be seen immediately, without having to be developed. Sometimes, also, it gives more distinct dots and so it is perhaps better when the print is to be studied under the microscope for special purposes. The print remains legible for several weeks at least. If it is needed for longer it must be photographed. A photostat copy is sometimes sufficient, but some detail is lost. Nevertheless, the iodine-starch prints photograph better than the ninhydrin prints.

The great advantage of the ninhydrin method lies in the fact that the print can be fixed and preserved. This makes it possible to compare prints taken at different times in their original form. This is valuable both for scientific work and for clinical purposes. For this reason I have used the ninhydrin method more often in this study. Otherwise, in my experience, the methods are of about equal value, and therefore they will be considered together henceforth.

As mentioned, the epidermis of the skin also contains substances that stain with ninhydrin. Consequently, when moist paper is used, intense prints are obtained even from a surface which has lost all power of secretion. These stainable substances probably result from the disintegration of proteins in the surface of the skin. From a theoretical point of view, it would be interesting to know whether the results of the ninhydrin method are dependent

mainly on the excreted amino acids, and not on the secretion of water as are those of the iodine-starch method. In such a case the methods would differ in principle.

I made some attempts to wash out the amino acids from the skin to see if this were so. A normal subject was made to perspire freely with salicylic acid and warm tea. When one of his finger pulps was washed with soap and water for twenty-five periods of two to ten minutes and tested between times, the ninhydrin prints became much weaker but they never disappeared. With every washing they took on more the character of punctiform prints of the sweat gland orifices. Washing with ether, alcohol and acetone had the same effect. This is a strong indication that the stainable substances were directly excreted from the glands. It does not seem likely, even though their corkscrew paths through the horny layer are long, that the epidermis could continually give off new amounts of stainable substances from the walls of the paths to the water in the sweat. The probability is, therefore, that the ninhydrin test registers the excreted amino acids, while the idoine-starch test registers the water excreted. So far, this has not meant any practical difference between the two methods.

RELATIONSHIP BETWEEN SENSIBILITY AND SUDOMOTOR FUNCTION

Recent nerve injury with total loss of sensibility in corresponding region—When a peripheral nerve is severed, the area of skin that it supplies stops sweating almost at once and has certainly done so by the time the patient reaches a doctor. On examination it is then seen that the regions with intact sensory function sweat normally (perhaps a little more than usual) and give normal prints, but the denervated regions leave no print at all. Figure 10 gives a clear illustration of this. It shows the case of a hand with the median nerve severed, resulting in loss of sensibility on the volar surface of the thumb, index finger, middle finger and partly on the ring finger. Another case is seen in Figure 11. There the thumb and little finger show normal sensibility while the ring finger has lost its sensibility because of the injury at the base of the finger. The prints show this clearly. There is no doubt about the relationship in these cases.

Experiments with nerve block—In order to study how the loss and return of the sensibility were related to the sudomotor function, the nerve was put out of function temporarily in a number of cases. The median nerve was blocked at the wrist in nine cases, and the ulnar nerve at the elbow in seven. The brachial plexus was anaesthetised in ten cases. The subjects were of both sexes and varied in age from sixteen upwards. None of them had any nerve injury.

In some cases the experiments were done in connection with an operation, in other cases not. They were continued until full sensibility had returned. The nerve block was produced with xylocaine in a 2 per cent solution to which epinephrine was added in the proportion of 1 : 80,000. A comparison was made between the sudomotor function, registered in the manner previously described, and the results of the customary tests of cutaneous sensibility.

The results being essentially the same in all the cases, I shall only give an example of each kind.

Blocking of median nerve—The prints are seen in Figures 12 to 15. The subject was a man of thirty-five years of age with a normal right arm. No premedication was used. The room temperature was 20 degrees centigrade. No operation was done. The prints of the finger pulps were normal (Fig. 12). The median nerve was blocked at wrist level with xylocaine.

Thirty minutes after the injection there was total anaesthesia and analgesia in the median nerve area. Thumb opposition was lost. The iodine-starch and ninhydrin tests (Fig. 13) showed that the area supplied by the median nerve had stopped sweating entirely, but that the area supplied by the ulnar nerve perspired normally, as well as the area on the lateral surfaces of the thumb innervated as border zones by the radial nerve. Repeated testing gave the same result until four hours after the injection, when thumb opposition began to return.

Five and a half hours after the injection pinpricks and touch began to be perceived in the anaesthetised region. Return of the sudomotor function was also manifested in the form of twelve dots in the print from the pulp of the thumb, one from the index finger and eight from the middle finger (Fig. 14).

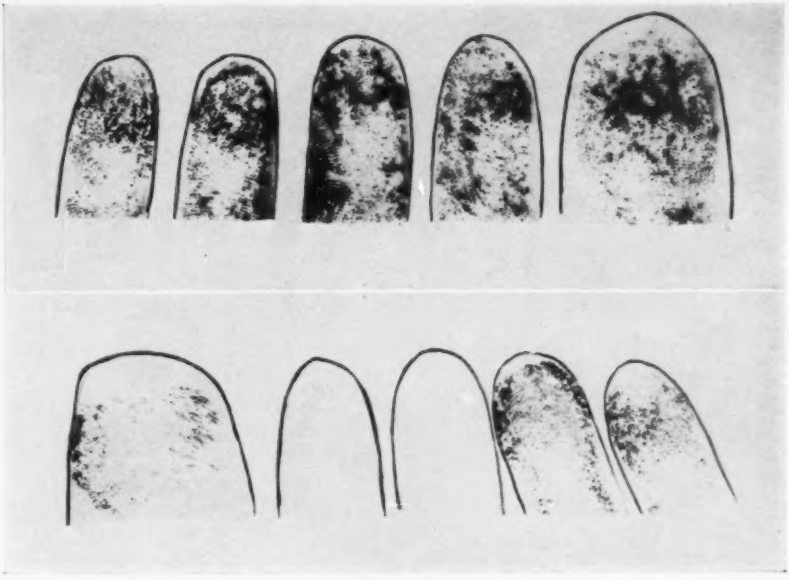

FIG. 10

Ninhydrin printing test. Median nerve severed at wrist level. Pulp of the thumb rolled from side to side over paper to include lateral surfaces in the examination. Dorso-lateral borders of thumb distally innervated by radial nerve. The normal hand is shown above for comparison.

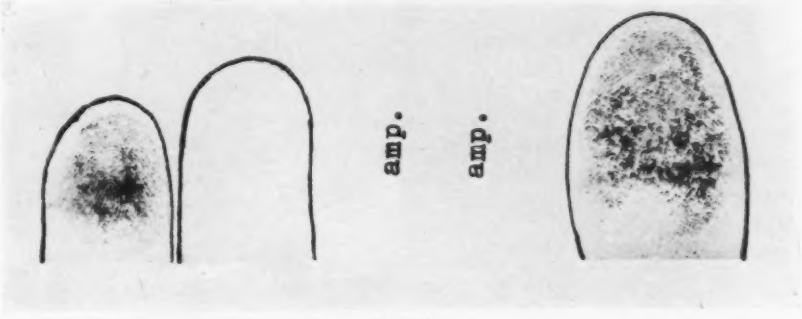

FIG. 11

Ninhydrin printing test. Thumb and little finger show normal sensibility. Index and middle fingers lost. Ring finger has no tactile gnosis, because of an injury at the base of the finger, where both digital nerves were severed.

Six and a half hours after the injection all that was left of the anaesthesia was a slightly numb feeling in the index finger. The sensibility was normal except that the two-point discrimination was about two or three millimetres worse in the index finger than in the little finger. The sudomotor tests gave normal results (Fig. 15).

Blocking of ulnar nerve—The same arm as in the previous experiment was used four days later (Figs. 16 to 19). The same experimental conditions prevailed. The initial fingerprints were normal (Fig. 16). The effect on the motor function will be disregarded here. The ulnar nerve was blocked with xylocaine.

Twenty-five minutes after the injection the involved region was completely insensible (Fig. 17). Repeated testing showed no change until three and a half hours after the injection,

FIG. 12

FIG 13.

FIG. 14

FIG. 15

Median nerve blocked at wrist level with xylocaine. Figure 12—Normal prints before injection. Figure 13—Thirty minutes after injection. Loss of tactile gnosis in median nerve area. Thumb rolled over paper to show remaining sensibility in border zones supplied by radial nerve. Figure 14—Three and a half hours after injection. Function has begun to return. Figure 15—Six and a half hours after injection. Full function restored.

when the sensibility began to return. The analgesia was then no longer sufficient for an operation. The sudomotor function began to return but the dots were still much fewer than in the median nerve region (Fig. 18). From then on both the sensibility and sudomotor function returned rapidly.

Four hours and twenty minutes after the injection all that remained was slight numbness in the ulnar nerve region. Points seven to eight millimetres apart were distinguished in the

pulp of the little finger as against four millimetres apart in the middle finger, but no other difference could be found. By this time the sudomotor function was almost normal again (Fig. 19).

Brachial plexus anaesthesia—This form of anaesthesia was used for ten similar experiments. In these cases a minor operation was done in the hand in a bloodless field produced by a pneumatic tourniquet on the upper arm. This did not appear to affect the results in any way.

FIG. 16

FIG. 17

FIG. 18

FIG. 19

Ulnar nerve blocked at elbow. Figure 16—Normal prints before injection. Figure 17—Twenty-five minutes after injection. Tactile gnosis has disappeared in ulnar nerve area. Figure 18—Three and a half hours after injection. Function has begun to return. Figure 19—Four hours and twenty minutes after injection. Almost normal function restored.

The results were essentially the same as when the median and the ulnar nerve were blocked. In some cases the sensibility returned first to the area supplied by the median nerve and in others to the area supplied by the ulnar nerve. The first signs of restoration of sensibility always coincided with the first signs of restoration of the sweat gland function. It took about an hour from these first signs until the function was almost completely restored.

These experiments show clearly how the sensibility and sudomotor function are interrelated. As soon as the first dots appeared in the prints, some capacity to perceive pain

TABLE I

COMPARISON BETWEEN GRIP FUNCTION, TWO-POINT DISCRIMINATION AND SUDOMOTOR FUNCTION

Case number	Age		Interval between injury and nerve repair (*months*)	Interval between nerve repair and follow-up examination (*months*)	Protective sensibility
	at injury (*years*)	at examination (*years*)			
1	36	41	0 (primary)	51	+
2	13	16	12	24	+
3	22	30	21	67	+
4	23	26	8	22	+
5	9	9	4	0 (before repair)	+
6	55	57	8	19	+
7	18	18	6	0 (before repair)	Thumb + Index − Middle −
8	14	18	48	0 (before repair)	Thumb + Index − Middle −
9	28	28	9	0 (before repair)	−
10	28	66	No repair	38 (after injury)	+

Comments: In Case 7 all sensory branches in the palm were severed; in the others the median nerve was divided at wrist level or in the middle of the

returned. When the sudomotor function was almost restored, the two-point discrimination began to approach the normal. Only a slight subjective feeling of numbness, hardly possible to register objectively, then remained for a few hours, the length of time varying from case to case.

Accordingly, these methods of registering the sweat-gland function provide an exact and objective means of following the variations in the sensibility of the hand produced by nerve block in the arm.

This is probably the only method by which the effect and duration of a local anaesthetic agent can be measured objectively.

COMPARISON BETWEEN TACTILE GNOSIS AND SUDOMOTOR FUNCTION AFTER LONG-STANDING MEDIAN NERVE INJURIES

In my aforementioned investigation I compared the results of the customary methods of examining sensibility with the actual functional usefulness of the same sensibility in ten cases. Here I shall compare the results of the objective printing methods in the same ten cases with the results of the two-point discrimination test, the only one of the subjective methods useful for judging the functional value of hand sensibility and for which the results can be given in figures. The results of this are seen in Table I and Figure 20. Normal sudomotor function is indicated by 3, moderately reduced by 2, greatly reduced by 1, and absent by 0.

It is clear that the better the tactile gnosis the denser are the dots from the orifices of the sweat glands in the prints. The presence of a few dots in a print does not mean that the area possesses useful sensibility. When about half the normal number of sweat glands leave an imprint, some tactile gnosis is present, and as the number of dots increases so does the tactile gnosis of the area. This makes it possible, for example, to examine the return of tactile gnosis

TABLE I—*continued*

Grip function			Two-point discrimination in pulp (*millimetres*)			Sudomotor function		
Gross grip	Precision-sensory grip	Picking-up test	Index	Middle	Thumb	Index	Middle	Thumb
+	+	+	8	15	5	2	1	2
+	−	−	20	20	15	1	2	2
+	+	+	15	12	12	2	1	2
+	+	+	12	10	12	1	2	2
+	−	−	>20	>20	>20	0	0	1
+	−	−	>40	>40	>40	1	0	1
+	−	−	>60	>60	15	0	0	2
−	−	−	>40	>40	>40	0	0	2
−	−	−	>60	>60	>40	0	0	1
+	−	−	>40	>40	>40	0	0	1

forearm (Case 10). No other lesion of functional importance. Case 10 was one of gunshot wound, the others of cuts by knives or glass.

after nerve suture, which is not possible with Guttmann's quinizarin method. As seen from the table, the two-point discrimination test and printing tests agree about the condition of the grip sensibility. A person with normal sudomotor function can perform the picking-up test, though perhaps not with normal speed, and distinguish two points down to twelve or fifteen millimetres apart. When the sudomotor function is absent, on the other hand, two-point discrimination is always impaired and the precision-sensory grip is lost. There is some evidence that the sudomotor function returns a few months before the tactile gnosis, but this is not yet established.

An apparent exception is sometimes found in the thumb. Thus in Case 8 the thumb possessed a fair sudomotor function (grade 2) but poor two-point discrimination. Case 6 showed similar conditions. These exceptions are only apparent, however. When the dorsal nerves of the thumb are blocked dorsally at the base of the distal phalanx with xylocaine the sudomotor function disappears at once. Closer study shows that in these cases it is mainly the lateral narrow border zones volarly on the distal phalanx of the thumb which leave prints of the sudomotor function. However, these narrow borders are so small that it is difficult or impossible to examine the tactile gnosis there with Weber's compasses, and so this method is apt to be misleading. The coin test often gives a clear verdict about the tactile gnosis in these small border zones (Fig. 10). Figure 20 shows a few examples of how the prints look after blocking the dorsal nerves to these zones. Then the results agree entirely with those of two-point discrimination.

Accordingly, Weber's compass test is apt to be unreliable in detailed examinations of narrow areas, especially when the testing can only be made along one line and cannot be checked by putting the legs of the compass at right angles to their first position. However, the prints give reliable results in these cases. This is important, because in a case of median nerve injury at wrist level the border zones are often what provide the thumb with good

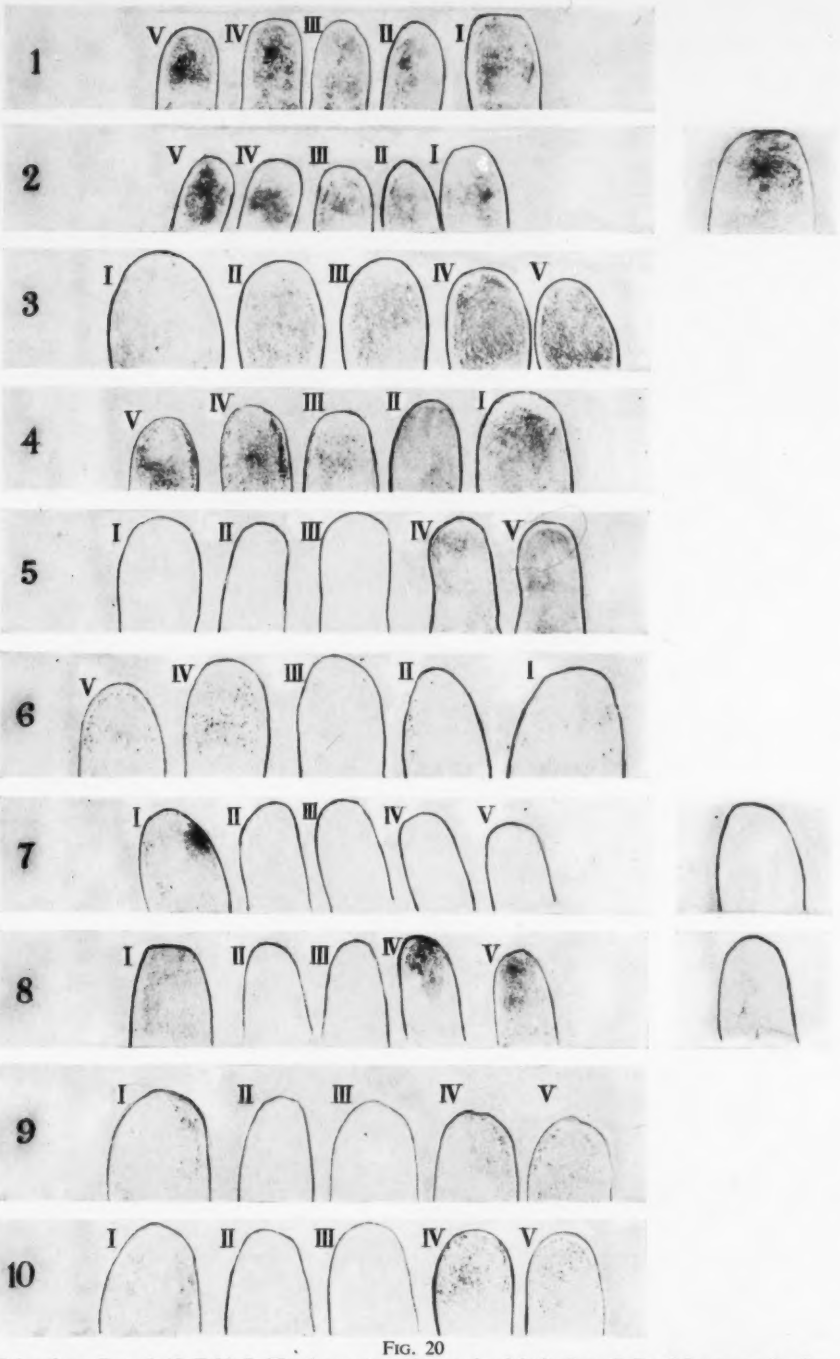

FIG. 20

Prints from Cases 1–10, Table I. The three extra prints at the right in Cases 2, 7 and 8 were made after blocking the radial nerve branches to the thumb. The prints are difficult to reproduce photographically without losing some of the details because the fixing solution shades the paper strips pale green.

tactile gnosis and make it useful for gripping with the ring and little fingers. When the border zones are insensitive the functional value of the thumb is greatly reduced. If the zones are broad the thumb may remain almost as useful as it was before it was injured.

DISCUSSION

It has been said (Guttmann 1940, Zachary and Roaf 1954) that loss of the sudomotor function is generally, but not always, accompanied by loss of sensibility. They found most exceptions in the trunk and face. These authors made little mention of different kinds of sensibility. In my experience, the loss of tactile gnosis in the hand is invariably accompanied by the loss of sudomotor function and *vice versa*, and the borderline between normal and abnormal areas can always be determined as exactly as it is possible to do with any known testing method.

It is of no use, when judging hand function, to divide the regions of nerve distribution in the ordinary way into autonomous and intermediary or to test separate physiological modalities. The so-called compensatory activity of nerve fibres from neighbouring, normally innervated zones occurring early after denervation does not provide tactile gnosis. For this there must be real nerve regeneration through the main trunk of the region. In none of the aforementioned cases without real nerve regeneration did the area lacking tactile gnosis distinctly decrease from what would correspond to the region normally innervated by the injured nerve. An illustrative case is Case 10, in which tactile gnosis was completely lacking in the whole median nerve area thirty-eight years after the denervation. It makes no difference even when a skin region is doubly innervated, as in the case of the tip of the thumb. After loss of one of the sources of innervation in these areas, what remains of the tactile gnosis undergoes no change unless repair is done.

Thus, in the hand, the regions which possess sensibility of the kind providing tactile gnosis coincide with the ones which have complete or almost complete sudomotor function. So far as is known, this parallelism is based on a purely anatomical coincidence, namely, that these two nerve elements follow the same pathway.

Comparison between the best of the subjective methods—the test of two-point discrimination—and the printing methods shows the following.

1. When the subjective factors are reduced to a minimum—for example, with a skilful examiner and the same co-operative experimental subject—differences can be demonstrated with the two-point discrimination test that are beyond the power of the printing methods in their present form. This is true, for example, of the differences normally present between the pulps of the index and little fingers, between the pulp of the finger and the volar area of the basal phalanx, and between the latter area and the palm. In all these instances, the two areas differ by a few millimetres in their ability to distinguish between two contacts.

2. Only the printing methods can reliably demonstrate small degrees of residual function in a greatly damaged nerve trunk. Only they, likewise, can be relied upon for testing narrow areas.

3. The boundaries between regions with and without tactile gnosis can be drawn more precisely with the printing methods, especially when tactile discrimination falls to ten millimetres of separation or more.

4. When the tactile gnosis is reduced in a region that may be supplied by two nerves, their relative share in the function can be demonstrated with nerve block and prints much more exactly than by any subjective method. This is sometimes useful to know when an operation is planned.

5. The greatest advantages of the printing methods are that they are completely objective and that the original test results can be kept.

To sum up: the tactile gnosis in the hand can be demonstrated by examining the sudomotor function in the same region with the ninhydrin and iodine-starch printing methods. When the sudomotor function is defective, a corresponding defect in tactile gnosis is present.

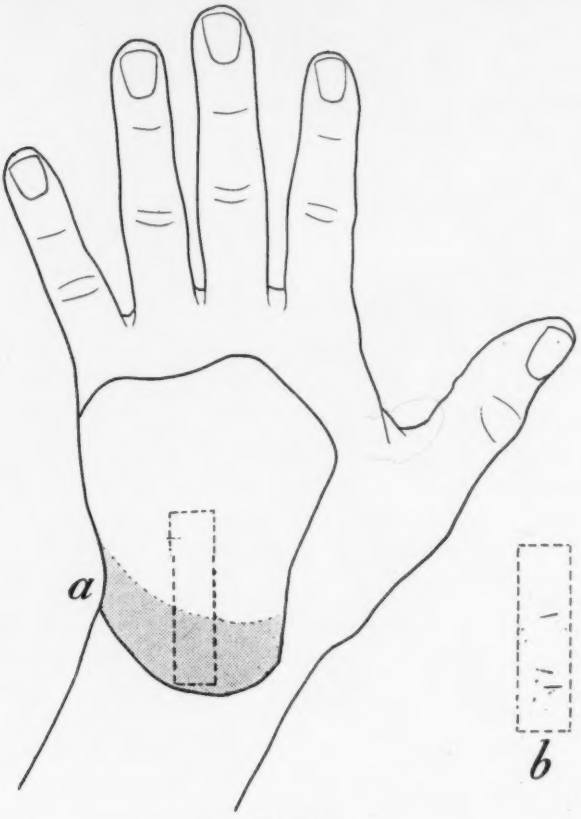

FIG. 21

Some sudomotor function returned to a skin graft in fourteen months, but tactile gnosis did not return.

Man aged thirty-five. Electrical burn. Two and a half months later, scar excision. Graft from thigh; thickness about three-quarters of the corium. No important nerve injury. Fourteen months later the sensibility around the graft was normal. Thus touch was perceived with the estesiometer on 0·5 gramme pressure and the two-point discrimination was normal. The graft contained a narrow zone (*a*) where 1·5 grammes were perceived, but otherwise it showed little sensibility. Heavy touch was perceived and likewise the estesiometer on 2·5 grammes pressure. Two-point discrimination was lacking. Distinct sweat gland function, however, had returned in the proximal part of the graft, as was shown by coarse clumsy prints (*b*) (indicated by interrupted lines). The function disappeared on plexus anaesthesia, as did the sweat gland function in the normal skin beside the graft, but it returned to both places when the anaesthesia disappeared.

The exceptions to this rule are few and quite distinct, and cause no great trouble. It has already been mentioned that the parallel ceases when the plexus is injured proximal to the site of entry of the sympathetic post-ganglionic fibres in the plexus. Likewise, operations on, or injuries to, the sympathetic nervous system in the cervical region may distort the results.

In addition to these obvious exceptions, however, there is one other which is of great interest both from a practical and theoretical point of view.

Exception to rule—The sudomotor function comes back to skin transplants but not the tactile gnosis. Many believe that sensibility comes back to all transplants after one or two years, both to so-called intermediary grafts and whole-skin grafts and flaps possessing subcutaneous

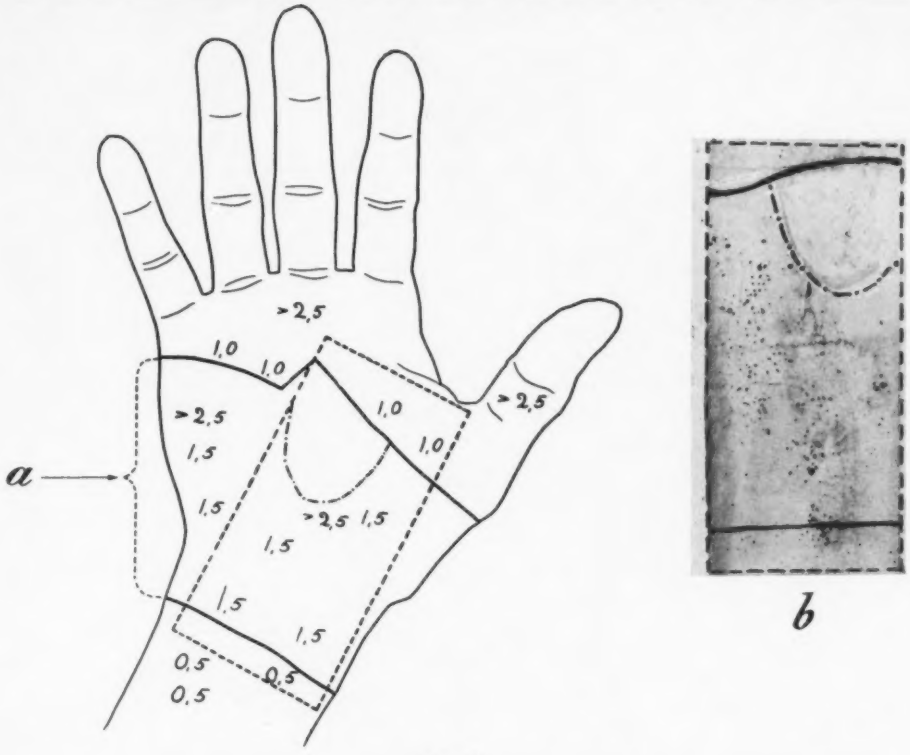

FIG. 22

Sudomotor function, but not tactile gnosis, returned to an abdominal flap in seventeen months.

The same patient as in Figure 21, also had a deep burn on the front of the wrist and palm of his other hand. Many centimetres were lost of both the median and ulnar nerves and of several flexor tendons. After excision of a deep scar and operations on the nerves and tendons the defect was covered with an abdominal flap (*a*). This flap was released from the abdomen seventeen months before examination. The final size of the flap was 11 × 12 centimetres. At examination the two-point discrimination was 10–12 millimetres in the skin proximal to the flap, and the sensibility there was normal in every respect. Two-point discrimination was lacking in the flap. Distally to this it was present (about 20 millimetres) in a narrow zone and, distally to this, lacking again. Estesiometer values are shown in the figure. The algesiometer needle caused pain of atypical character on 1 gramme pressure in the small zone distal to the flap. Distal to this no pain was perceived with loads up to 10 grammes, and the same was true of the whole flap. Sweat gland function proximal to the flap was also normal. Distal to the flap it was lacking. The flap itself had distinctly functioning sweat glands, though they gave coarse and clumsy prints (*b*) compared with normal skin and with the abdominal area from which the flap was taken. The function disappeared during plexus anaesthesia but returned with the normal sweat secretion of the skin proximal to the flap when the effect of the nerve block disappeared. Only a small area distally in the flap, where no sensibility at all was observed, gave no prints. Distal to the flap where the skin was left *in situ* there were still no signs of sweat gland function.

fat. That they regain sweat gland function has been shown by Löfgren (1951) and others. The return of sensibility to skin grafts has been studied by Napier (1952) and others. Napier observed that three different types of reinnervation took place, and made a careful analysis of the anatomical factors that determined the forms in which the sensibility returned. He did not study the returning sensibility by means of two-point discrimination, though he used this

method elsewhere in the same work. It is easy to deduce why: two-point discrimination does not return in the skin graft, at least to any useful degree.

I compared the sensibility with the sudomotor function with all the usual methods and with the printing methods in seven skin transplants which had been in place for a long time. Essentially the same results were obtained in all of them—namely, returning sudomotor function but no tactile gnosis (Figs. 21 to 23). A number of grafts that had been in place for only three or four months were also tested and proved to lack the sweat gland function and all sensibility. Moreover, several other whole skin grafts and flaps which had been in place for up to nine years were tested only for sudomotor function and two-point discrimination. All proved to lack the degree of discrimination necessary for tactile gnosis, though they possessed the sweat gland function. Among these were grafts that were transferred from one part of the hand to another, as cross-finger flaps, and thus consisted of skin which before the transfer had good tactile gnosis.

These observations show that skin grafts transferred to the hand do not regain tactile gnosis. Thus in every case in which the graft was so situated that its usefulness for a precision-sensory grip could be tested, the power of tactile gnosis was lacking and the picking-up test was a failure.

It is interesting, however, that the sudomotor function returned. In skin grafts, accordingly, it does not vary parallelly with the tactile gnosis. It may even return to tissue in which protective sensibility is still lacking, as in the case shown in Figure 23.

The sweat glands, however, did not function in the normal way. The prints were not those of the delicate glandular openings in normal skin, even on the abdomen. Instead, they consisted of blurred and irregular spots, as if a large amount of sweat had collected in a dilated channel and gushed out all at once.

A fact of great theoretical interest is that skin which is denervated by nerve injury but still normally connected with the surrounding tissue does not acquire the same sudomotor function as transplanted skin. Both may acquire about the same capacity to register touch and pain. Both lose the power to distinguish between two separate points of contact, but this capacity can only come back to skin *in situ* and only after successful nerve repair. The sweating function which returns to the transplant is controlled by the nerves for, in my cases, it disappeared when the brachial plexus was blocked.

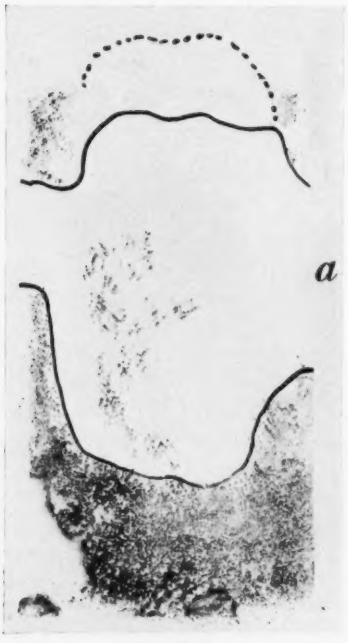

FIG. 23

FIG. 23

Some sudomotor function, but not tactile gnosis, returned to a flap in ten months. Ninhydrin printing test. Man aged thirty-nine. Avulsion injury with loss of the skin from dorsum of hand. The area was covered with an abdominal flap 9 × 10 centimetres in size. The flap was detached ten months before the examination. The flap (*a*) showed practically no form of sensibility. Only rough handling like lifting a fold of tissue with a pair of forceps was perceived. There was no protective sensibility. Proximal to the flap and on both sides the sensibility was normal. Distal to the flap, however, was a denervated region about 3 centimetres wide, which showed no two-point discrimination and did not react to 10 grammes load on the algesiometer needle or 2·5 grammes pressure with the estesiometer. Proximal to the flap the sweat gland function was normal. Distal to it, in the anaesthetic zone, it was lacking. The flap itself showed distinct sweating in the form of clumsy spots in the prints. This secretion, too, disappeared when the brachial plexus was blocked.

An explanation for this might be that new nerve fibres to the denervated sweat glands follow along with the new vessels which grow into the graft. Denervated skin *in situ* which does not acquire new vessels would not have the same chance to have its sweat glands reinnervated.

To summarise: tactile gnosis does not return to skin grafts but the sudomotor function usually returns after a year or so. Apart from what has been said before, this is the only exception to the rule that in the hand the tactile gnosis varies with the sudomotor function. The exception is so clear cut that it should not cause any difficulty.

PROCEDURE FOR DETERMINING THE FUNCTIONAL VALUE OF THE CUTANEOUS SENSIBILITY IN A HAND

A hand or part of a hand that lacks tactile gnosis, even if it possesses one or more of the aforementioned modalities of sensibility, is nevertheless " blind "—that is, it cannot be used without the aid of the eye; it does not know whether or how it holds an object, or what the object is. Only by determining the tactile gnosis can a real idea be obtained of the value of the cutaneous sensibility for the precision-sensory grip.

The functional value of the cutaneous sensibility in a hand, therefore, should be determined in the following way.

Tactile gnosis for precision-sensory grip (Fig. 3)—This can be determined objectively with the ninhydrin or iodine-starch printing methods. Subjective methods that may be used as a complement are the two-point discrimination test, Seddon's coin test and the picking-up test.

Sensibility necessary for gross grip (Fig. 4)—The presence of this function can be determined objectively by looking for callosities and similar signs of usage in the hand, and subjectively by testing the hand in use.

Protective sensibility—This must be judged from the history unless typical lesions show that it is absent.

The methods routinely used hitherto for examining the sensibility to touch and pain in a hand must be reserved for the neurological purposes mentioned, where they are of proved value.

EXAMPLES OF THE PRACTICAL APPLICATION OF THE METHODS

1. Guttmann has already shown with the quinizarin test that the regions supplied by the various nerves vary from hand to hand. The methods described here are much more exact, simpler to perform and are not objectionable for the patient. They permit detailed study of the nerve distribution.

2. Their value for study of local anaesthetics has already been discussed.

3. Hitherto there has been no objective method for judging disability in the sensibility in the hand. The customary methods, all subjective, are highly unsatisfactory and result in unfair financial consequences. The simple methods described here give the information desired. It is easy to keep records of the prints.

4. When there is an acute injury involving essential nerves in the hand, particularly the median and ulnar nerves, mistakes are often made when the hand is first treated. Because of the injured person's mental state after the injury, it is easy to understand that serious mistakes are made with subjective methods, mistakes that have a grave effect on the future earning capacity of the patient. Errors of this kind can be avoided by using the objective diagnostic methods (naturally before any nerve block for anaesthesia).

5. In difficult elective cases important diagnostic details can be clarified before operation much more exactly than with the other more time-consuming methods. The following cases illustrate this.

Case 1—A woman aged sixty-one suffered from nerve compression in the carpal tunnel caused by chronic non-specific tenosynovitis. For about six months she had noticed a swelling at the front of both wrists, much greater on the right side. Her fingers sometimes felt numb and clumsy—for instance, when she raised a cup to her mouth, buttoned her clothes or knitted. The only objective signs were a swelling over the front of the carpus, weakness and slight atrophy of the thenar muscles, and tactile discrimination of about six millimetres in the pulp of the right index finger as compared with three to four millimetres in the index finger of the other hand and in the ulnar-innervated fingers. The ninhydrin method, combined with dorsal block of the radial branches, showed that the sudomotor function in the median nerve region had deteriorated as compared to the normal hand. Operation disclosed typical chronic non-specific tenosynovitis compressing the nerve in the carpal tunnel.

In this case, since it was possible to verify the change in sensibility objectively, the hand was operated upon early, while the nerve lesion was still reversible.

Case 2—A girl of eighteen had cut the palm of her right hand with glass six months before she came to us. It was then seen that the thenar muscles were still innervated. This did not necessarily mean, however, that the motor function of the median nerve remained, but may have been due to an innervation of most of the muscles by the ulnar nerve. The sensibility was reduced but not absent in the pulp of the thumb. The same was true of the ring and little fingers. It was lost entirely in the volar surface of the index and middle fingers. It was known that the injury must lie near the branching area of the median nerve in the palm, which meant a difficult dissection in scar tissue. As the sensibility was already so much reduced in this hand, it was important not to injure any branch where there was still some useful nerve continuity or where regeneration had started. Yet waiting meant taking the risk that nerve suture would have to be done at a later, much more unfavourable, date.

Good prints were obtained with the ninhydrin method, showing tactile gnosis in the pulp of the thumb and a small amount in the ring and little fingers. It was also found that the dorsal skin of the ring and little fingers had retained its capacity to perspire even beyond the middle phalanges. This was surprising, for the region supplied by the dorsal nerves usually ends at the proximal interphalangeal joints. There was a possibility that it meant that the volar nerves still functioned to some extent. However, on dorsal block of the nerve branches on the thumb, little finger and ring finger, the perspiration stopped entirely, both volarly and dorsally. This established that all the volar nerve branches must have been cut off and that the volar sensibility in both the thumb and in the ring and little fingers must have been derived from the dorsal branches of the radial and ulnar nerves respectively. No subjective methods would have been able to establish with such certainty that this unusual anatomical anomaly was present. Operation completely verified the pre-operative diagnosis.

6. The methods may provide the only means of making a correct prognosis at an early stage of nerve injury, as the following case shows.

Case 3—A man aged thirty-seven had sustained a crushing injury of the upper arm, with a transverse wound ten centimetres above the elbow on the inner aspect of the arm. There were all the signs of severe injury to the median nerve with loss, perhaps total, of motor function and sensibility, and also an incomplete lesion of the ulnar nerve. Immediate testing with the ninhydrin method (Figs. 24 and 25) showed about fifteen sweat gland openings in the pulp of the index finger, reduction of the sudomotor function in the middle finger and great reduction of this function in the thumb. The perspiration in the thumb and middle finger might have been explained by anomalous innervation. However, it was hardly possible so to explain the function in the index finger: it must have meant that there was nerve conduction past the site of the lesion. Because of this, the outlook was pronounced favourable. A week later the print shown in Figure 25 was obtained. With subjective methods it was hardly possible to demonstrate any sensibility coming from the median nerve in the thumb and index finger, and the motor function was still almost completely lacking.

Six months later the prints were almost normal. The sensibility was then normal in the middle finger. Points ten to twenty millimetres apart could be distinguished in the pulp of the index finger and twelve to fourteen millimetres apart in the pulp of the thumb. There was already considerable motor regeneration and it was still in progress.

Thus, it was possible with this method to exclude complete interruption of the nerve on the day of the accident. With other methods this could not have been done until months later.

Theoretically it should be possible to use the printing methods for distinguishing between pre-ganglionic and post-ganglionic injuries of the plexus. In such a case they could be used instead of the much more complicated "cold dilation test" (Bonney 1954) used for this purpose. This would make it easier to foretell the outcome of a severe plexus injury at an

FIG. 24

FIG. 25

Case 3—Crushing injury above elbow with open wound. No unquestionable median nerve function could be demonstrated with the customary methods. However, the ninhydrin printing test revealed nerve function even in the index finger, and as this could hardly have been due to an anatomical variation, the prognosis was pronounced favourable at the first examination (Fig. 24). A week later the test showed improvement (Fig. 25), and a few months later good, but not normal, function was restored.

early stage and eliminate the long period of expectancy before starting on reconstruction. I have not had access to any cases suitable for study of this kind.

7. These methods can be used in many ways for planning reconstruction of the grip. A number of these possibilities will be demonstrated in another paper.

SUMMARY

1. It was observed clinically that tactile gnosis varies directly with the sudomotor function in the hand.

2. Two methods of fingerprinting were elaborated to register the sudomotor function, and consequently the tactile gnosis objectively. They are sensitive, simple to perform and suitable for clinical work. Their anatomical background, sources of error and relative value are discussed.

3. The correspondence between the sudomotor function, determined with these methods, and the tactile gnosis was established. This was done by, firstly, comparing the regions which did not perspire with the ones which became insensible on total denervation of a region of the hand; secondly, by examining the loss of function after nerve block; and thirdly, by comparing the tactile gnosis and sudomotor function in cases of residual median nerve defect.

4. These two qualities do not accompany each other in skin grafts. Grafts regain sudomotor function but never tactile gnosis.

5. A practical procedure for determining the functional value of the cutaneous sensibility in the hand is described.

6. Cases are related illustrating the usefulness of objective study of the sensibility in the hand.

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TWO CASES OF OBSTETRICAL SEPARATION (EPIPHYSIOLYSIS) OF THE UPPER FEMORAL EPIPHYSIS

Appearance of Ossification Centre of the Femoral Head in a Fifteen-day-old Child

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Obstetrical fractures (epiphysiolysis) of the upper epiphysis of the femur are among the rarest and most interesting injuries in the new-born. About forty cases have been reported, but it is probable that many such cases have been overlooked because many obstetricians, radiologists and paediatricians are insufficiently aware of the signs of this fracture. Early diagnosis is at first a clinical one, as radiological signs become evident only after an interval.

Although some authors (Madsen 1955) have claimed that they have discerned " as early as the third day a faint callus shadow," this seems to be very uncommon: we have never seen a callus shadow at such an early date. Radiological signs may, on the other hand, be misinterpreted as showing congenital dislocation of the hip (Nathan 1928, Puppel 1930). A careful clinical examination as well as an accurate history of the delivery will eliminate this diagnostic error. So far as we are aware the existence of an obstetrical (traumatic) dislocation of the hip has never been demonstrated. Experiments (Riedel 1956) have shown, on the contrary, that if violent traction is exerted on the leg of a still-born infant, an epiphysiolysis but not a dislocation of the hip will result.

The first reported cases of an obstetrical separation of the upper femoral epiphysis seem to be those of Deesfo (1855) and Pforte (1908), but the most complete studies of this injury are those of Camera (1929) and Harrenstein (1929).

In most cases delivery has been from breech or foot presentations. The clinical symptoms are characteristic: the new-born child lies with the limb in slight lateral rotation and adduction, avoiding and resenting movement. Swelling of the thigh is constantly found. Radiographs taken immediately after birth may show a displacement of the upper end of the femur, which must never be confused with a congenital dislocation of the hip. The displacement is often slight and is therefore difficult to interpret. Later the radiographic sign that gives irrefutable evidence of the existence of an obstetrical fracture is calcification of the haematoma. Although Brailsford (1953) asserted that this calcification is visible in the first week after birth, we have never seen it distinctly before the third week.

In our first case the ossific nucleus of the femoral head appeared at the early age of fifteen days. This enabled us to diagnose with certainty the separation of the upper femoral epiphysis, because the head remained in its normal position in the acetabulum, whereas the shaft was displaced upwards. It is worth emphasising that in this case the ossification centre became visible before the callus shadow was apparent. So far as we know, there is no record of the appearance of this centre of ossification before the age of two months.

The usual mechanism of the injury is one of hyperextension, abduction and rotation during strong traction on the leg as it is brought forward: the separation occurs at the growth cartilage. In the new-born this lies below the greater trochanter, extending in an arrow-like line to the lesser trochanter (Trueta 1957). A circular periosteal detachment, which may sometimes involve the whole surface of the femoral shaft, occurs simultaneously.

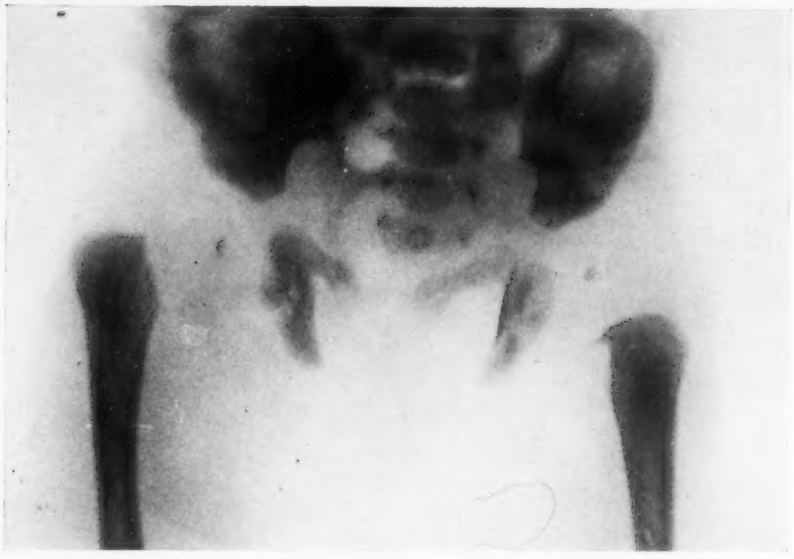

FIG. 1

Case 1—Radiograph nineteen days after birth. Marked elevation of the right femoral shaft, which might be mistaken for a congenital dislocation. The ossification centre of the femoral head is already visible in the acetabulum, thus eliminating this possible diagnostic error.

FIG. 2

Case 1—Radiograph thirty-eight days after birth, showing profuse callus formation about the upper end of the femur. The ossification centre is much larger.

CASE REPORTS

Case 1—Girl, forty days old when admitted. She was born after a difficult delivery. The obstetrician had exerted strong traction on the right leg. The parents, anxious about the condition of their child, visited many doctors before attending our clinic. On admission, we noted a bulky right thigh, and the limb lay in adduction, lateral rotation and slight flexion. The limb was in a fixed position and any attempt to move it appeared to cause pain. Radiographs taken when the child was fifteen days old showed the ossification centre of the femoral head, which was the size of a pin head. This remained in its normal position in the acetabulum; the shaft was displaced in such a way that it suggested a congenital dislocation of the hip. With this unexpectedly early appearance of the ossification centre, the diagnosis of the fracture, instead of a congenital dislocation of the hip, was undoubted. A second

FIG. 3
Case 1—Five months after birth. Marked coxa vara.

radiograph taken on the nineteenth day after birth (Fig. 1), showed that the ossification centre had doubled in size. Nevertheless no shadow of callus formation was visible either on the fifteenth day or on the nineteenth day. It would have been interesting to have another radiograph before that taken on the fortieth day after birth. In this latter film (Fig. 2), the callus was well marked but in the meantime the size of the ossification centre had greatly increased. Five months after birth there was marked coxa vara (Fig. 3). No later follow-up has been possible.

Case 2—Girl, aged three months when admitted. She was born in a private clinic. Breech presentation. Delivery was difficult. The right thigh was noted to be bulky. The new-born child kept her limb immobile, protesting whenever the parents attempted to change this fixed position. A radiograph taken a week after birth was considered normal (Fig. 4) and no treatment was therefore given. Much more attention was paid to an obstetrical palsy of the right arm. Radiographs taken on admission (Fig. 5) showed still no ossification centre in the right acetabulum (nor in the left), but a bulky metaphysis surrounded by spindle-shaped

FIG. 4

Case 2—Radiograph taken a week after birth. Though the child had obvious clinical symptoms of an epiphysiolysis no evidence of it was seen in the radiograph. The upward displacement of the left femur (which was not injured) was surprising: this is ascribed to congenital subluxation.

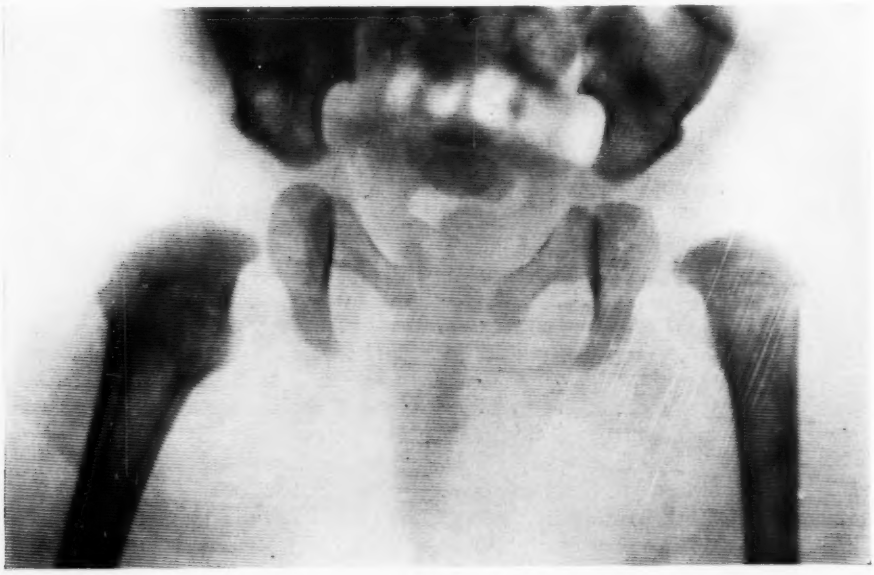

FIG. 5

Radiograph taken on admission three months after birth. The right upper epiphysis is bulky and there is subperiosteal ossification of the upper part of the femoral shaft. The ossification centre of the head of the femur is not visible.

FIG. 6
Case 2—Radiograph six months after birth. Note subluxation of the uninjured (*left*) hip.

FIG. 7
Case 2—Radiograph eighteen months after birth. Almost normal appearance of both hips.

callus. There was also thickening of the periosteum of the shaft. Curiously enough, a congenital subluxation of the left hip was to all appearances present on the left side. No violence had been exerted on the left lower limb. This interesting observation supports our opinion that a violent traction causes a fracture rather than a dislocation of the hip.

Later radiographs (Figs. 6 and 7) showed progressive improvement in the appearance of the upper end of the femur, which after eighteen months became nearly normal.

DISCUSSION

We present these two cases because we believe that they will be of interest to orthopaedic surgeons as well as to obstetricians, paediatricians and radiologists. The obstetrician should be extremely careful in his manipulations in difficult deliveries. He has to remember, especially in breech presentations, the possibility of an epiphysiolysis which is a serious injury.

When the diagnosis has been made—mainly on clinical grounds—treatment should be started as soon as possible. The application of vertical traction, followed, if necessary, by immobilisation in plaster, seems to be the most adequate procedure. Manipulative reduction has also been recommended (Alcivar 1946). In our two cases the patients were referred to us too late to allow satisfactory treatment. Nevertheless the result in the second case was good because the leg had been secured in a proper position.

SUMMARY

1. Two new cases of obstetrical separation of the upper femoral epiphysis are described.
2. Diagnosis in the first instance must be largely clinical, because radiological confirmation of the injury may be delayed.
3. The simple method of longitudinal traction is the treatment recommended.
4. It is notable that in our first case the ossification centre of the femoral head appeared at the exceptionally early age of fifteen days.

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FAMILIAL CONGENITAL POSTERIOR DISLOCATION OF BOTH RADIAL HEADS

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Congenital dislocation of the head of the radius is not common. Bilateral posterior dislocation appears to be very rare.

CASE REPORT

A young Yoruba farmer brought his second child, a girl, to University College Hospital, Ibadan, when she was six days old because movement of her elbows had been noticed to be limited since her birth. Birth had been by a normal spontaneous vertex delivery and the child was otherwise healthy, but neither elbow could be extended below the right angle.

FIG. 1
Left arm of daughter at fullest extension. The dimples were present on both sides.

Pronation and supination were unrestricted. The baby's subcutaneous fat prevented accurate localisation of the radial heads by palpation, but the elbows were visibly deformed (Fig. 1) and radiographs showed backward dislocation of the heads of both radii (Figs. 2 and 3).

The father's elbows could also be extended no farther than the right angle. All other movements in the upper limb were full, including pronation and supination. The tendons of insertion of both biceps stood out prominently in his antecubital fossae, raising skin webs (Fig. 6). His triceps were poorly developed, but all other muscles appeared normal. The heads of the radii could be readily felt in abnormal posterior positions, as illustrated in the radiographs (Figs. 4 and 5). The deformity was symmetrical. The radii were longer than normal and there was no constriction to mark their necks, the heads and the shafts being of equal girth. The radial heads were not dome shaped. Despite the good development of his biceps the bicipital tuberosities were hypoplastic and not clearly seen in the radiographs.

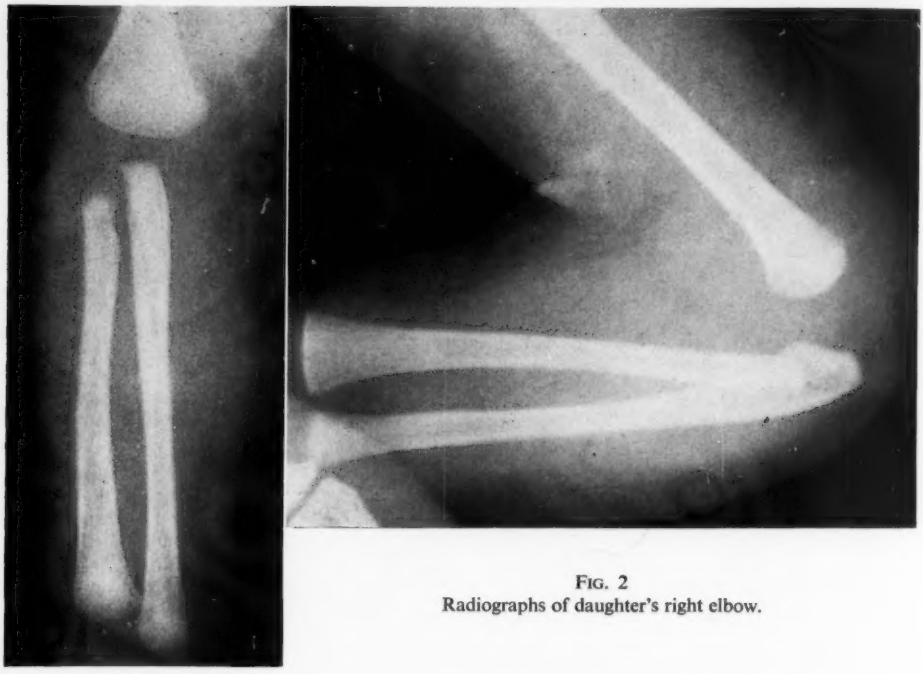

FIG. 2
Radiographs of daughter's right elbow.

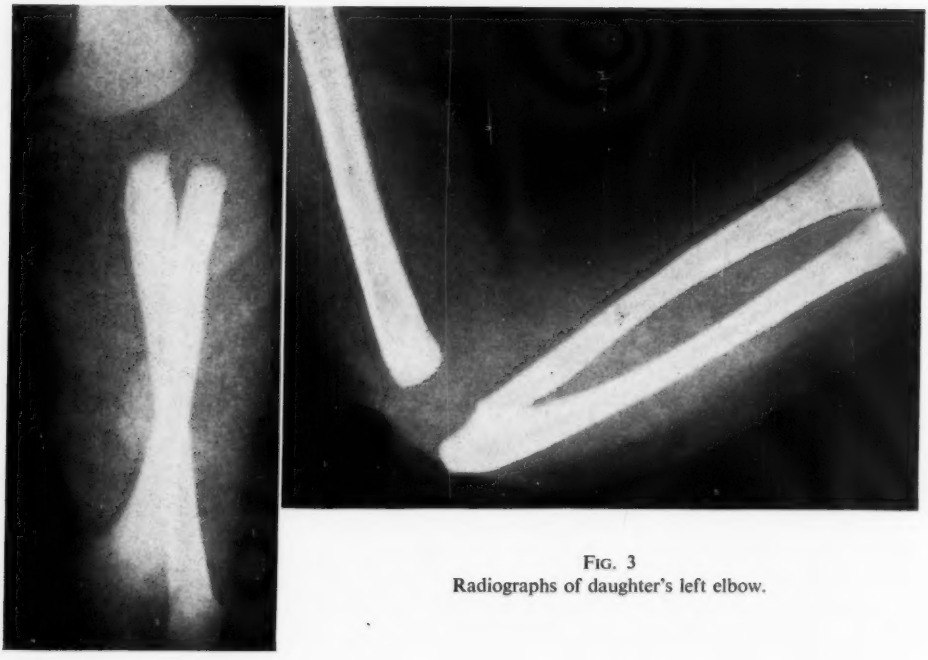

FIG. 3
Radiographs of daughter's left elbow.

FIG. 4
Radiographs of father's right elbow.

FIG. 5
Radiographs of father's left elbow.

The region that is normally the neck of the radius appeared to be touching the lower aspect of the capitulum, causing a bony block to extension. The medial humeral epicondyles were large and were situated proximal to their usual positions. Limitation of extension of the elbows had been present from birth, but it caused very little disability except for hoeing, which he was unable to perform in the native manner.

We were not able to trace any other examples of this deformity in the family. The baby's elder sibling, a boy, had lived only five days. His elbows were reputedly normal. The father came of a large family, but none of his relatives (except his daughter) was affected.

FIG. 6

Left arm of father showing greatest degree of extension of the elbow. The biceps tendon is prominent anteriorly.

DISCUSSION

Abbott (1892) reported seven cases of congenital anterior dislocation of the head of the radius in one family. McFarland (1936) described the anatomy of the anterior congenital dislocation of the radial head. Both anterior and posterior dislocations may occur in association with gross congenital abnormalities such as radio-ulnar synostosis or arthrogryposis multiplex, but we have found no report of bilateral posterior congenital dislocation in otherwise normal persons, nor of congenital dislocation of the radial head in father and daughter.

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UNEXPLAINED DESTRUCTION OF THE SHAFT OF THE FEMUR IN A CHILD

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A five-year-old boy was admitted to an isolation hospital in Kent in August 1947 under suspicion of poliomyelitis. Although he showed no evidence of acute illness, he had had a slight limp, with apparent weakness of the right thigh muscles for the previous few days. In the past he had been free from all serious illness, but four months previously he had had his tonsils removed, and within a few days of leaving hospital had developed a mild attack of measles from which he recovered rapidly. Thereafter he was well for over three months before the limp appeared.

On examination the only abnormality found was slight weakness of the quadriceps and hip flexor muscles on the right side, and since at that time there seemed to be no local cause it was thought likely that he had had a mild attack of poliomyelitis before entering hospital. Lumbar puncture was not performed. After a period of physiotherapy he returned to his home and resumed school, still limping a little on the right leg but complaining of no discomfort. A month later, while he was at play, the shaft of the right femur broke at the junction of the uppermost and middle thirds. The fracture was treated by traction in the children's ward of a general hospital. It gradually became clear that this fracture was no straightforward traumatic lesion. After the initial formation of a small amount of callus on the lateral side of the femoral shaft all further progress towards union ceased. The callus reabsorbed, and in the course of the next five months the greater part of the uppermost and middle thirds of the shaft of the femur disappeared. Throughout this period, and no doubt from at least the time of onset of the limp, it is clear that some locally destructive process was at work in the femur; but, although investigations were carried out when the failure of the fracture to repair normally was appreciated, no satisfactory explanation of this was forthcoming. Apart from pain at the time of the fracture there was no local discomfort or swelling of the limb, no upset of general health and no pyrexia or other evidence of general or local infection. He had no anaemia or leucocytosis. Blood calcium, phosphorus and phosphatase were normal. The Wassermann and Kahn reactions were negative.

In January 1948, as no other cause of the local bone lesion had been found, it was thought possible that there might be an underlying upset of metabolism producing osteomalacia with a spontaneous fracture, though the rest of the skeleton, judging by subsequent radiological survey, was in no way affected and the local lesion in the right femur did not suggest the type of pseudo-fracture often associated with osteomalacia. In January and February 1948, and again in May and June 1948, full courses of vitamin D therapy were therefore given, being stopped in each case when the blood calcium rose above normal, and on the second occasion when this was accompanied by toxic symptoms of headaches and vomiting. There appears to be no evidence that this vitamin D therapy affected in any way the course of the lesion in the femur.

The destructive process in the femur progressed steadily until at least the end of March 1948, eight months after the original onset of symptoms. Thereafter it appears to have come to an end spontaneously, leaving the femur still ununited and, in spite of splintage by a plaster spica, with much shortening.

Radiological findings—In August 1947, before he returned from hospital to his home and school, and four weeks after the onset of the limp, radiographs were taken of the hips and right knee.

Seemingly, through some error, the abnormalities revealed were not recorded before the boy left hospital, but the films were found many months later and were then available for comparison with those taken after the fracture had occurred. In the radiographs taken to show the hip joints much of the shafts of both femora were also shown (Fig. 1). In the uppermost and middle thirds of the femoral shaft the medial cortex appears to be greatly thinned and in some areas has practically disappeared. The shaft also appears narrowed, as if the medial cortex had been absorbed, partly at least by pressure from without, the contour of the medial surface being concave.

The radiograph taken a month later, at the time of the fracture (Fig. 2), showed that considerable further bone destruction had occurred on the medial and posterior aspects of the shaft; and that taken six weeks later still (Fig. 3), when there was slight callus formation

FIG. 1
Initial radiograph showing early changes in the shaft of the right femur.

on the lateral side of the bone, shows that destruction had passed right across to the lateral cortex near the site of fracture. The appearance is more like that of a malignant neoplasm than of any infective process.

Further radiographs taken from January to March 1948 (Fig. 4) show that the uppermost third of the shaft became almost completely destroyed, and in the middle third only the lateral cortex survived. After March the osteoporosis of the bone on either side of the area of destruction was so intense that it is impossible to see detail in the films or to reproduce them satisfactorily. It seems, however, that after the beginning of April 1948 no more actual bone destruction occurred, and in the next eight months the bone remaining in the upper two-thirds of the shaft slowly became better calcified.

By the end of 1948 the right femur was represented by a somewhat porotic proximal fragment consisting of the head, neck and trochanteric region (Fig. 5), tilted into a flexion contracture of some 40 degrees with slight abduction at the hip joint. Distal to this, two small separate fragments represent all that was left of the uppermost third of the shaft down

to the site of fracture. Below these, and separated from them by a slight gap, the proximal end of an upwardly tapering process represented the lateral side of the middle third of the shaft which had survived destruction. This merged into the lowermost third of the shaft which was almost normal in shape but poorly calcified except for the juxta-epiphysial zone where the newly formed bone showed better density.

This was the state of affairs in January 1949 when the boy was transferred to Addenbrooke's Hospital under the care of Professor R. A. McCance.

FIG. 2

FIG. 3

FIG. 4

Figure 2—Six weeks after onset of limp. Spontaneous fracture of femoral shaft. Figure 3—Six weeks later. Some attempt at union of fracture but further bone destruction proceeding at the same time. Figure 4—Six months after onset. Disappearance of most of the uppermost and middle thirds of the femoral shaft. Osteoporosis was so marked that the films have had to be " touched up " to make reproduction possible.

A full clinical and biochemical investigation showed no general or local abnormality apart from the state of the right femur. A radiological survey showed all other bones to be normal. In the course of the next few weeks it became clear that the destructive process was at an end, and the patient was transferred to the Orthopaedic Department as presenting a problem of bone loss requiring reconstructive surgery.

Treatment—The interest of the case is essentially the unexplained nature of the destructive process in the femur which progressed for at least nine months and then came to an end equally mysteriously. The subsequent operative treatment, which eventually resulted in restoration of continuity in the shaft of the femur, is therefore only of special interest in that it provided an opportunity for exploration of the site of the lesion and for biopsy and

histological examination and culture of bone immediately adjacent to the area of destruction. Apart from these points the treatment is, therefore, described only briefly.

In June 1949 an operation was carried out to explore the site of the lesion, to restore as much length as possible and to bridge the affected area by a rigid bone strut combined with autogenous cancellous bone grafts. The site of the lesion was exposed by a long anterior incision between the rectus femoris medially and the vastus lateralis and tensor fasciae latae

FIG. 5

FIG. 6

FIG. 7

Figure 5—A year and a half after onset. State of right femur after cessation of destruction and some recalcification. Figure 6—Right femur four years after onset (two years after operation). Figure 7—Ten years after onset (eight years after operation).

muscles laterally. In the area of discontinuity the original line of the shaft was represented by a fibrous strand linking the bone fragments and suggesting a remnant of the periosteum. There was no scar tissue between the muscles, no macroscopical evidence of past inflammation and no abnormal soft tissue of any kind.

Samples of bone were taken from the tip of the upward tapering end of the lower fragment and from the small fragments above this. Some of the smaller fragments were cultured and found to be sterile.

Little faith was placed in the attenuated remnant of the middle third of the shaft as a source of osteogenesis, and further operation was planned as a virtual replacement of the

upper two-thirds of the shaft. The space representing the upper two-thirds of the shaft was, therefore, bridged by a nine-inch rigid strut of boiled dried human tibial cortical bone, sharpened at both ends (Fig. 8). The upper end of this was impacted into the trochanteric region of the upper fragment and the lower end into the comparatively normal-sized lowest third of the shaft.

Autogenous cancellous bone, not available in quantity from the ilium of a child, had been obtained at the start of the operation by removing subperiosteally about six inches in each case of the fifth, seventh and ninth ribs on the right side. The three six-inch segments of rib were split longitudinally and were placed, like a bundle of sticks, on the front and lateral side of the rigid strut, overlapping the main upper and lower fragments at either end. Most of these were held in place by loops of stainless steel wire (Fig. 8D). About two inches of shortening of the femur were accepted. The limb was then immobilised in a plaster spica.

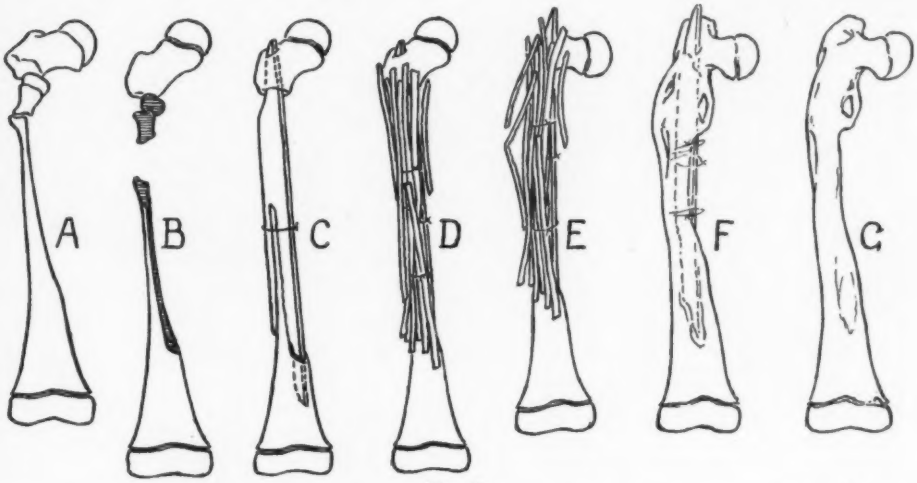

FIG. 8

Diagrams of operative procedure on June 1, 1949, and its result. A. The femur before operation. B. Lengthening obtained at operation. Shaded areas of bone removed for culture and microscopy. C. Insertion of rigid strut of boiled tibial cortical bone. D. Application of strips of cancellous bone from split ribs. E. Breaking out of upper end of rigid strut from porotic upper femoral fragment with recurrence of deformity and shortening five weeks after operation. F. Restoration of continuity by new bone spreading through cancellous grafts. Rigid cortical strut is unincorporated in live bone and acting as a sequestrum, shortly before its removal in May 1950. G. State of restored femoral shaft in October 1950.

About a month after the operation the upper end of the rigid tibial graft broke out of the porotic trochanteric region, and the upper fragment again tilted into flexion with some varus deformity (Fig. 8E). Nevertheless, though some extra shortening resulted, in the course of the next six months a bridge of new bone replaced the bundle of cancellous bone grafts and restored continuity between the upper and lower ends of the femur. Ten months after operation the rigid tibial graft which had remained unincorporated in live bone, and which was acting as an irritant with the formation of a sinus which became indolently infected, was removed (Fig. 8F, G). From that time onwards the femoral shaft thus restored grew steadily in bulk and strength (Fig. 6).

Presumably as a result of the very long periods of splintage in plaster and by traction, the trauma of operation, and perhaps also from damage inflicted by the original destructive process, the epiphyses of the lower end of the femur and also of the upper end of the tibia began to fuse prematurely from about midsummer of 1950. This produced progressive comparative shortening of the leg and a marked genu valgum. The latter was corrected by

an osteotomy in May 1953. In June 1953, when the child was only eleven (but for his age already long in the normal left leg) epiphysiodeses of the left lower femoral and upper tibial epiphyses were carried out. In January 1957, aged sixteen, he was seven inches short in the right leg; but, wearing an O'Connor type extension shoe and a long leg caliper which stabilised the limb for walking, he was of comparatively normal appearance when wearing long trousers, and was working in comfort as a junior clerk in a local authority's office (Fig. 7).

Histology—Sections of the upper end of the lower fragment are illustrated in Figures 9 to 11. The free upper end of the bone in Figure 9 terminates in cellular connective tissue. The rest of the bone, and the whole of that in Figure 10, consists of compact bone of abnormal architecture. Part of this—for instance, the upper part of the right side in Figure 10—has

FIG. 9 FIG. 10 FIG. 11

Figures 9 and 10 show sections of the long upward tapering extremity of the lower part of the femur: Figure 9 shows the apex, and Figure 10 the bone immediately below and continuous with it. Figure 11 is part of the same section as Figure 10, at higher magnification, to show the large thin-walled blood vessels in the endosteal connective tissue. (Celloidin sections: Ehrlich's haematoxylin and eosin. Figures 9 and 10, ×5; Figure 11, ×50.)

the general pattern of callus; and the presence of abundant islands of woven bone in this part confirms that it has been newly formed. The rest is of somewhat looser structure and does not contain woven bone; it consists of long parallel seams of lamellar bone separated by moderately cellular fibrous connective tissue in which there are numerous thin-walled blood vessels. Some of the spaces in Figure 10 are artefacts, but Figure 11 shows a representative sample of the vascular endosteal tissue at a higher magnification, so that the blood vessels can be clearly seen. Those shown are of average size; many others are larger and more cavernous. Some of these abut on the surface of bony trabeculae, and there appears to be only endothelium between blood and bone. The outer surfaces of the lamellar bone are mostly smooth and covered by an uninterrupted layer of osteoblasts which in some places is more than one cell thick. Osteoclasts and Howship's lacunae are present focally but are not numerous.

The cap of the cellular connective tissue (Fig. 9, *top*) consists of spindle-shaped cells resembling fibroblasts, amongst which there are some eosinophil leucocytes, but no foamy macrophages or other chronic inflammatory cells such as are usual in eosinophilic granuloma. A moderate number of small round cells and very few macrophages and plasma cells are present in parts of the endosteal connective tissue.

COMMENT

From the clinical and radiological evidence at our disposal and the late and inconclusive biopsy no firm diagnosis of the cause of this widespread local bone destruction can be made. Furthermore, our knowledge that the boy was apparently well throughout and that the destructive process came to an end spontaneously eliminates several otherwise feasible explanations.

An eosinophilic granuloma, affecting a single bone, is a theoretical possibility. The upper part of the shaft of the femur is a recognised site for eosinophilic granuloma, and their radiological appearances are notoriously variable. An isolated white cell count in December 1947 showed 12 per cent of eosinophils, but unfortunately no further white cell counts were made until nine months later (September 1948) when the eosinophil cell count was found to be normal. Although eosinophil leucocytes are present in the connective tissue in the biopsy, the histological findings as a whole are, however, far from characteristic of eosinophilic granuloma.

The abundant thin-walled blood vessels present in the histological sections are of interest. This vascularity may perhaps be merely the normal vascularity of young connective tissue, but if so it might be expected to be greatest in the evidently younger callus-like parts of the specimen, and this is not so; furthermore, the cavernous appearance of some of the vessels is noteworthy. On the evidence available, however, we would not feel justified in suggesting that the picture is that of an angiomatous condition, so that both the cause of this vascularity and its relationship to the previous disappearance of bone remain unknown.

MASSIVE OSTEOLYSIS—DISAPPEARING BONES

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This is the record of a case in which there was complete disappearance of the left scapula and the outer third of the clavicle and partial absorption of several ribs. Death finally occurred from chylothorax due to obstruction of the thoracic duct by fibrous tissue.

Twenty-six such cases have been recorded before, but only one in England (Jackman 1939). Opportunities for complete pathological study have been rare, although a fair amount of biopsy material has been available. From the literature it appears that one other patient died as a direct result of the disease, and in that case also chylothorax occurred after disappearance of the right clavicle, most of the scapula and several ribs (Gorham, Wright, Shultz and Maxon 1954). These authors studied two cases of their own and sixteen others from the literature.

The condition was the subject of a further review, with full references, by Gorham and Stout in 1955, when the total number of cases tabulated was twenty-four. Gorham and Stout's account is particularly important because the authors studied the original sections from a total of eight patients and concluded that on histological grounds there was a common, basic pathological lesion. Different authors have described the histological changes as " connective tissue and capillaries," " fibrous tissue rich in blood vessels," " marrow replaced by fibrous vascular tissue," " resembles fibrocystic disease " and " abundant vascular channels." Gorham and Stout found a convincing similarity in the histological pictures seen in this condition, and stated that progressive osteolysis was always associated with angiomatosis of blood vessels, and sometimes of lymphatics, which seemingly was responsible for it. They were unable to state whether massive osteolysis occurred as the direct result of active hyperaemia, of mechanical causes, of slight changes in pH, or from some other cause. Since Gorham and Stout's report Krikler (1955) and Marie *et al.* (1956) have each added a further example.

It is clear that massive osteolysis or disappearing bone disease is quite distinct from atrophy after injury or from disuse. The osteolysis progresses until the affected bones are represented by fibrous tissue. It is unnecessary to repeat the details given in the full reviews mentioned above, but a few general points may be considered. The condition is most common in adolescents and young adults, and there is often a history of minor injury. The progress of the bone resorption is gradual, and is often accompanied by surprisingly little disturbance of function. Constitutional disturbance is usually slight, and there has been no significant alteration in the blood calcium, phosphorus, phosphatase or proteins when these have been recorded. Sometimes the disease has undergone spontaneous arrest: certainly there is no known effective treatment. It is not accompanied by any definite neurological abnormality.

CASE REPORT

The patient was a man born in 1928. The first record of trouble with the left shoulder was at the age of twenty-three, when he was admitted to a military hospital. The documents state that he was swinging three floor bumpers the previous day when the left arm gave way. It is also noted that three years previously he had " put out the left clavicle and went to a bone setter who put it back. Since then the left shoulder has always been weak." According to the notes, examination showed marked dropping of the left shoulder, the lower angle of the scapula being an inch lower on the left than on the right. There was marked wasting

of the supraspinatus and infraspinatus muscles and slight wasting of the deltoid. Marked wasting of the upper arm was noted. He was unable to raise the arm from the side or to maintain the passively abducted arm. There was no sensory loss, but he complained of acute tenderness over the scapula just below the spine. Radiographs are reported to have shown an apparent crack in the scapula about two inches below the neck. Unfortunately the original radiographs have been destroyed and only the reports remain. A report made one month after his first attendance noted an old united fracture of the left clavicle. A further report a year later indicated that the fracture of the neck of the scapula had healed. He was still having difficulty in raising the arm but there were no sensory changes; tenderness over the scapula persisted.

Records do not show when he left the Army, but the remark " deserted later " appears. It is recorded that he was at work in a quarry as a labourer for a year or more before he was

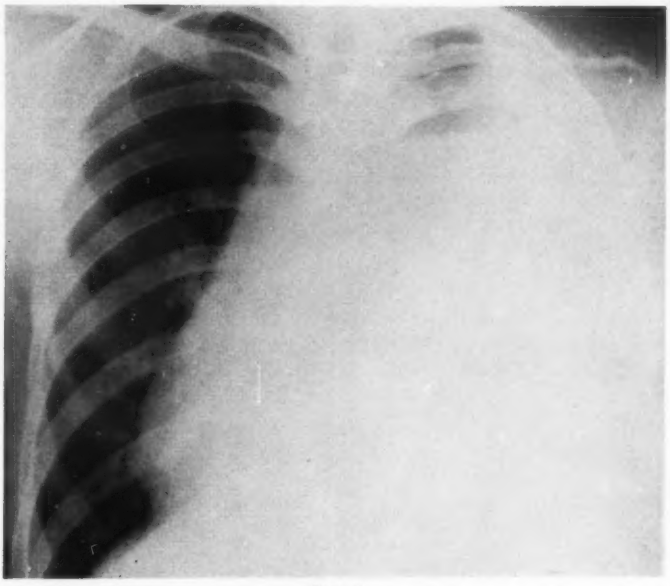

FIG. 1
Radiograph showing the large effusion in the left pleural cavity. The attenuated end of the left clavicle is also shown.

first seen, at the age of twenty-eight, by one of us (R. L. M.). He had noticed loss of weight for two years. He sought advice on account of pain in the left chest with shortness of breath and swelling under the left arm. He stated that the left shoulder had never been strong since onset of the trouble five years before, though he had been able to work quite well with it.

On his admission to Hawkmoor Chest Hospital he had marked dyspnoea, with a large chylous effusion in the left side of the chest (Fig. 1). There was a large fluctuant swelling in the left scapular area which could be compressed into the chest by pressure from the hand. There was considerable wasting of the left deltoid and spinati muscles; he was unable to raise the arm, though a limited range of passive movement was possible. There were no abnormal neurological signs. Chest aspirations were performed on twenty-six occasions on the left side during the ensuing month, a total of 45·6 litres of milky fluid being withdrawn. Towards the end of the month fluid began to accumulate in the right pleural cavity and seemed to diminish in the left. In the succeeding week three aspirations of the left pleural cavity

yielded 2·3 litres and three of the right pleural cavity 4·3 litres. Thus about 52·2 litres of chylous fluid were lost to the patient in forty days. This is equivalent to 116 lb., which was very nearly his body weight on admission to hospital.

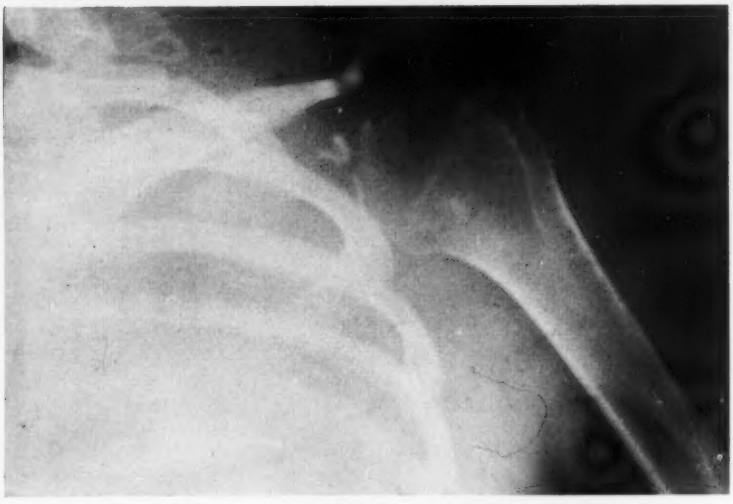

FIG. 2

Radiograph taken after removal of chylous fluid from the left pleural cavity. There is a faint shadow in the region of the glenoid cavity. The attenuation of the clavicle is well shown. There is nothing to indicate the position of the scapula.

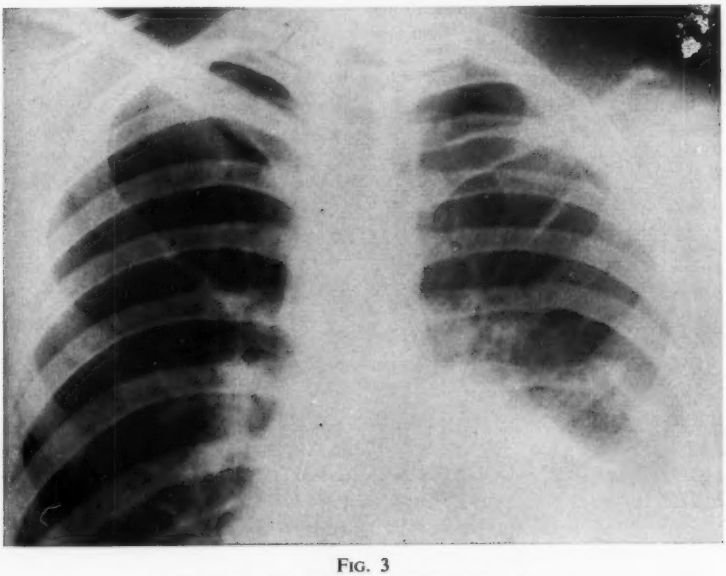

FIG. 3

Radiograph showing the erosion of the ribs.

When the fluid had been removed it became clear that he had no left scapula, and on palpation there was nothing to indicate where it should have been. Radiographs showed only a faint shadow in the position of the glenoid cavity and no evidence of any other part of

the bone. The outer half of the left clavicle had also largely disappeared (Fig. 2). There was erosion of the ribs in the position where the scapula should have been, and it was here that the communication existed between the chest wall and the pleural cavity (Fig. 3).

Investigations—Biopsy was done on the outer end of the left clavicle, and the histology of this is considered later. The pleural fluid was examined on several occasions for tubercle bacilli both culturally and biologically, with negative results. *Blood count* showed: haemoglobin 107 per cent; white blood cells 7,400 per cubic millimetre (polymorphonuclears 87 per cent, eosinophils 4 per cent, lymphocytes 8 per cent, monocytes 1 per cent). *Urine*—Bence Jones protein was not present. *Serum proteins*—Albumin 2·22 grammes per cent; alpha 1 globulin 0·35 gramme per cent; alpha 2 globulin 1·03 grammes per cent; beta globulin 0·83 gramme

FIG. 4

The head of the left humerus and the glenoid cavity. The glenoid cavity consisted of fibrous tissue and a little cartilage; outside this was some fibrous tissue but no evidence of a scapula.

per cent; gamma globulin 5·20 grammes per cent. *Fluid from the chest* showed large numbers of mononuclear cells but no acid-fast bacilli. Protein was 2·62 grammes per cent, and fat 1·105 grammes per cent. Fat globules were present after extraction with ether. The Wassermann and Kahn reactions were negative.

The patient died from cachexia seven weeks after his admission to hospital.

Post-mortem findings—The body was that of a greatly wasted man.

Skeletal abnormalities—The outer third of the left clavicle was missing, and in the fibrous tissue around the end of the clavicle there were two hard nodules probably containing bone. The part of the clavicle nearest this outer third was thin and tapered almost to a point. The shoulder girdle was dissected but no trace of the left scapula could be found, nor was there an excess of fibrous tissue in the position where the scapula might have been. A bunch of muscle was present but this did not appear to contain any bone or calcareous elements.

FIG. 5
Three of the left ribs. They were matted together at their angles: the bone was thinned and easily fractured.

FIG. 6
Tissue from the angle of an affected rib. Bone trabeculae separated by large amount of interstitial tissue containing large capillary channels. (H. and E., ×40.)

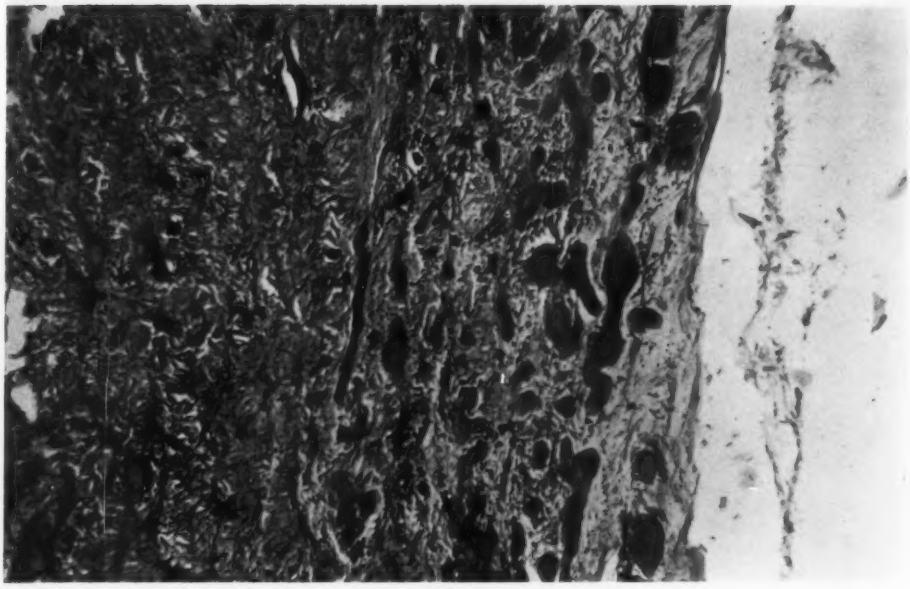

FIG. 7
Periosteum of rib and underlying fibrous tissue, showing large numbers of dilated capillaries. (H. and E., ×40.)

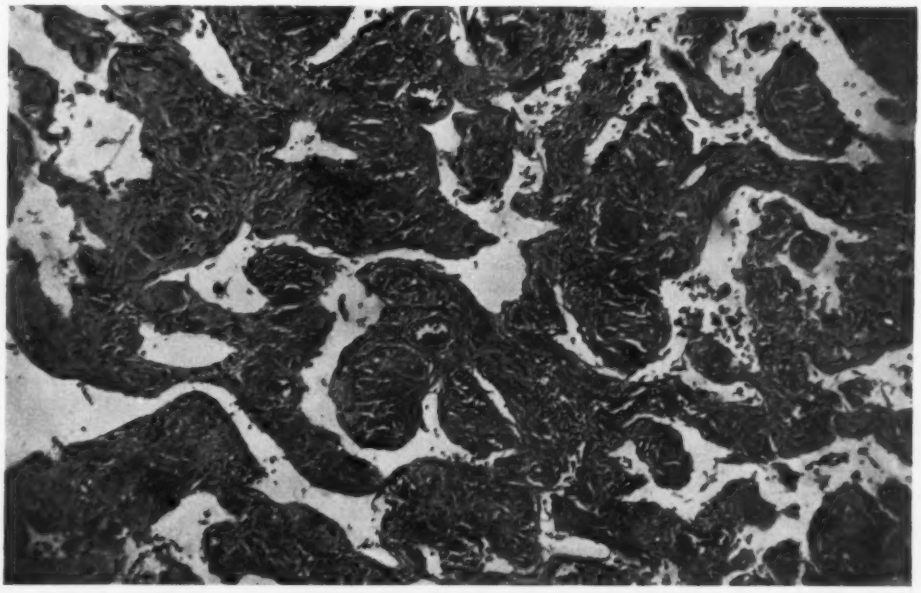

FIG. 8
Fibrous tissue from around the outer third of the clavicle, showing large anastomosing vascular spaces. (H. and E., ×80.)

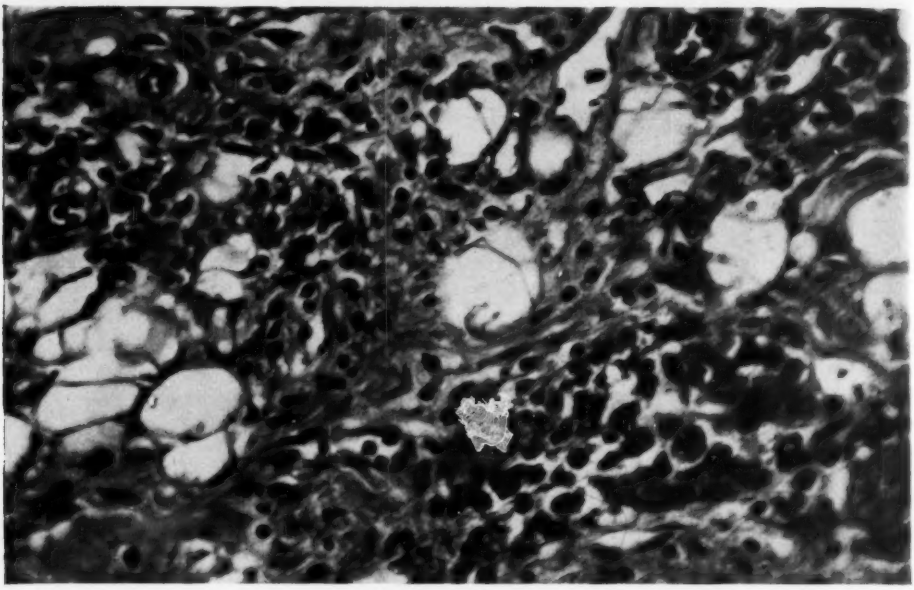

FIG. 9
Tissue from around the outer third of the clavicle. Plasma cells and eosinophils present in focal collections. (H. and E., ×320.)

It was difficult to imagine that there had been a scapula in this position and that it had disappeared entirely. The uppermost third of the left humerus was removed for further examination (Fig. 4). The head was well formed and of normal size, but on its inferior surface there was a considerable amount of erosion, leaving a rough corrugated surface. The capsule of the joint contained a little cartilage on the glenoid side but no bone. The ribs on the left side appeared to be normal anteriorly and laterally, but at their angles they were matted together. Three which were removed for examination showed that the bony portion of the rib became gradually thinner towards the neck of the bone (Fig. 5), and they were very easily fractured. When the anterior parts of the bodies of the vertebrae were removed the bony structure was normal.

Heart—The heart weighed 180 grammes. Though small, it was in keeping with the general physique. There was no valvular disease and the coronary vessels were healthy. The pericardial sac contained about four ounces of turbid, milky fluid. There was no excess of fibrin on the surface of the heart.

Lungs and pleural cavities—The right pleural cavity was full of turbid, milky fluid of chylous type and the lung was compressed. There was no evidence of neoplasm and the ribs appeared to be normal. On the left side, at the entrance to the chest cavity, surrounding the subclavian vessels, there was a mass of dense fibrous tissue which may well have obstructed the thoracic duct. This was removed for further examination. The lung was completely collapsed against the vertebral column, dark in colour and semi-solid, but there was nothing to suggest neoplasm. The chest cavity contained no fluid but did contain a large mass of fibrous material which was adherent both to the ribs and to the surface of the lung.

Other organs—The liver was normal in size and showed slight congestion. The spleen was about one and a half times the normal size: the cut surface appeared normal. The adrenals were normal. The kidneys showed no naked-eye evidence of disease. The alimentary tract showed no abnormality. The brain was not examined.

Histology—The tissues examined histologically included: 1) material obtained at operation from the outer third of the left clavicle; and 2) the remainder of the left clavicle, parts of ribs and the fibrous tissue surrounding the left subclavian vessels taken at necropsy.

The various bone specimens each showed a similar structure (Figs. 6 to 9). There was evidence of osteoclasis, the bone being replaced by fibrous tissue. The periosteum was thickened, and on its deeper surface it merged into fibrous tissue in which were many dilated capillaries. Similar large capillaries were present in the interstices of the bone. The degree of this vascular change was not even throughout the material. Evidence of new bone formation was scanty.

The material from around the subclavian vessels consisted of fibrous tissue containing large nerve bundles, sympathetic nerve ganglia and several large arteries and veins, some of which showed various stages of thrombosis and recanalisation. An interesting feature of this tissue was the presence of numbers of eosinophils and plasma cells in large groups, sometimes with a perivascular pattern. This feature was not found in the bone lesions. Small capillaries were not so prominent in this tissue, but large anastomosing vascular spaces, like those described by Gorham and Stout (1955), were much in evidence.

DISCUSSION

These clinical and pathological features appear to us to place this case in the rare group which shows massive osteolysis. The outer third of the clavicle and the whole of the scapula had disappeared, and portions of several ribs were in process of vanishing.

In particular, comparison of the histological findings with those described and fully illustrated by Gorham and Stout, in their study of eight examples of this condition, leaves no doubt that we are dealing with one of the very few cases in which the disease itself led to the death of the patient. The presence of eosinophils in the material from near the site of the clavicle raised the possibility of an eosinophilic granuloma, but further study of the vascular pattern, and especially of the reaction in the bones undergoing absorption, made it appear that this chronic inflammatory reaction was not the essential part of the disease process.

We do not feel able to suggest the etiological factor except to agree with Gorham and Stout that the osteolysis seems to be related to the haemangiomatous condition and may be a direct result of the consequent hyperaemia.

Injury was mentioned in the history of this patient, but it does not appear to us to be the cause of the condition. One can imagine an injury to the shoulder initiating a destructive lesion in the clavicle or the glenoid cavity, but complete disappearance of the scapula from this cause is most unlikely. Injury certainly cannot be the explanation of the osteolytic changes in the ribs. Trophic nerve lesions are not known to cause multiple foci of bone destruction of this magnitude.

SUMMARY

1. The clinical and pathological features are given of an example of the rare syndrome of progressive and massive osteolysis associated with haemangiomatous changes in the bones affected.

2. Twenty-six cases have been previously reported, but this is only the second from Britain. Only one other patient is considered to have died as a direct result of the disease.

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DISAPPEARING BONES

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There does not appear to have been any report in this country of a case of disappearing bones on the scale here presented, though a number of cases have been reported in the United States of America and on the Continent. The patient is an elderly woman in whom the bones of the left upper limb have been progressively disappearing.

Seemingly the first report of a case of this kind appeared in the *Boston Medical and Surgical Journal* in 1872, entitled " Absorption of the humerus after fracture "—the implications being that the fracture was the starting point of the condition. Possibly this is putting the cart before the horse.

Another case was reported by Branch in 1945, and a review of cases of vanishing bones was made by Gorham and Stout in 1955. They appended a bibliography of thirty-five items, and suggested that the " progressive osteolysis is always associated with an angiomatosis of blood and sometimes lymphatic vessels, which seemingly are responsible for it."

CASE REPORT

A single woman, born in 1887, complained of pain and incapacity in the left shoulder. She worked in a cotton mill until July 1947 when she gave up work to nurse a sick mother. She had enjoyed good health, but in 1941 had had her uterus removed together with a para-ovarian fibroid weighing 8 lb.

In 1945 she tripped and fell rather heavily on her left shoulder and arm, bruising the outer side from mid-arm to mid-forearm, but she did not stay away from work.

In May 1945, when consulting her doctor about cystitis, she complained of pain and stiffness in her left shoulder. Radiographs of the left shoulder showed an apparently normal humerus and ribs, but the whole of the clavicle and scapula presented irregular areas of absorption without any evidence of bone reaction (Fig. 1). She received a course of radiant heat and massage, with some improvement in the shoulder stiffness. No further notice was taken until two years later—December 1948—when radiographs to include the chest and both shoulders displayed the ribs still looking normal; but the humerus now showed gross rarefaction and all that remained of the clavicle and scapula was a bit of the lower part of each in the neighbourhood of the shoulder joint (Fig. 2). Seen a month later (January 1949) she complained of pain over the outer side of the left arm, the top of the shoulder and the scapular region, somewhat relieved by kaolin poultices. She had lost over a stone (14 lb.=6 kilograms) in weight in two years.

Examination—She had only a few degrees of movement in the left shoulder. She could get her left hand on to her right shoulder but little else. Movements in all the more distal joints were good.

The shoulder as a whole drooped, the thoracic cage giving a prominence in the mid-trapezius line, but there was no tumour mass. There was a nodule, about half an inch in diameter, in the right lobe of the thyroid gland.

Investigations—Haemoglobin 98 per cent. Red blood corpuscles 5,330,000 per cubic millimetre. Colour index 0·92. Mean diameter 6·5μ. White blood cells 5,300 per cubic millimetre (neutrophils 77·0 per cent; eosinophils 0·5 per cent; basophils 0·5 per cent; lymphocytes 17·0 per cent; monocytes 5·0 per cent). Inorganic phosphorus 3·8 milligrams per 100 millilitres serum (repeat 3·5). Alkaline phosphatase 21·5 units (repeat 25·5). Calcium 12·2 milligrams per 100 millilitres serum (repeat 11.0). Wassermann reaction negative. Serum proteins: albumin 4·0 per cent; globulin 4·2 per cent; total 8·2 per cent. Albumin/globulin ratio 0·96.

FIG. 1

FIG. 2

Figure 1—Initial radiograph, showing early involvement of the clavicle and scapula. Figure 2—Two years later. The clavicle has disappeared. Of the scapula, only the glenoid remains. Absorption of the humerus is well advanced.

FIG. 3

FIG. 4

Figure 3—Three and a half years after onset. The humeral head is merely a ghost. The spontaneous fracture of the surgical neck can be seen. Figure 4—Five years after onset. The upper half of the humerus has gone.

Stools—Total fat 35·3 per cent of dried faeces; split fat 87 per cent of total fat; unsplit fat 13 per cent of total fat. (These results indicated defective fat absorption with normal fat splitting.)
Control radiograph—A radiograph of one leg taken synchronously on the same film with the leg of another out-patient of like sex, age and build showed almost equal density in the tibiae of patient and control.
Arteriographs (Fig. 8) did not show any opacity such as might be expected with a haemangioma.
Biopsy—This was carried out in February 1949, through an anterior incision. It was difficult to recognise the anatomy, because the only bony point to be felt above the humerus was the

FIG. 5

FIG. 6

Figure 5—Eight years after onset. Only the lower end of the humerus remains. Figure 6—Ten years after onset. Further absorption of humerus, radius and ulna. To a less extent the hand was also affected.

anterior edge of the glenoid. The muscles arising from the coracoid process were traced upwards and found to be attached to a tiny fragment of bone having tenuous connections up into the neck. A finger put round the margin of the glenoid encountered a rough, rather sandpapery surface immediately behind the glenoid proper. The glenoid was removed by cutting round the capsule. This was sent for section. The remains of the coracoid were taken out, and the muscles were sewn to the remainder of the capsule around the head of the humerus.
Progress—In February 1950 the patient had increasing pain below the shoulder and she could not use the arm. Seen a week later, she presented a good deal of swelling around the lower

half of the humerus, and crepitus in the upper part of the shaft. Radiographs showed extreme rarefaction throughout the whole humerus; and there was a crack about an inch below the surgical neck, without displacement (Fig. 3). She was treated symptomatically with a light plaster slab.

In April 1950 the neck was explored by Professor John Morley, who removed the enlarged right thyroid lobe and a thyroid adenoma from the isthmus. The parathyroid glands were not seen. In the next year she gained 2½ stone (35 lb.=16 kilograms).

In May 1952 the upper end of the humerus had disappeared and there was nothing to be felt above its lowest third (Fig. 4). The whole arm was flail above the elbow. Further

FIG. 7

FIG. 8

Figure 7—Photograph of the patient, illustrating the flexibility of the humerus. Figure 8—Arteriograph (1958): no evidence of haemangioma.

radiographs taken in 1954 and 1957 are shown in Figures 5 and 6, and a clinical photograph is shown in Figure 7. The upper arm has become completely flexible and can be bent and twisted in any direction, so that the forearm can be made to assume all kinds of abnormal positions. The epicondyles can still be felt, and the ribs can be palpated in the upper part of the thoracic wall which normally lies under the scapula. The patient is free from pain. She has control of wrist and finger movements although she has nothing like normal power. Her general health remains good. She lives alone and does her own housework although she is now seventy. We have not been able to find evidence of bone disappearance in any other part of the skeleton.

PATHOLOGY

Pathology of biopsy specimen—The specimen consisted of part of the cartilaginous surface of the glenoid cavity with what little remained of the supporting bone. The sections showed extreme bone atrophy with small areas of active osteoclasis and numerous dilated thin-walled vessels. There was no evidence of tumour.

In the absence of any neoplastic or other evident cause of the bone absorption the case remained a mystery until reference to Coley's (1949) remarks on " Phantom Bone " revealed its identity.

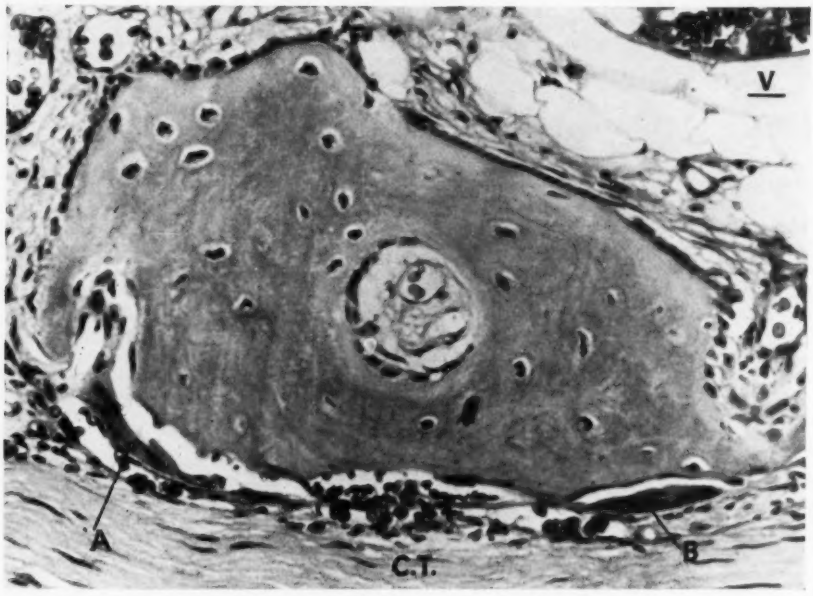

FIG. 9

Field from the deep surface of the cancellous bone below which is a connective-tissue layer (C.T.). The lower border of the bone shows an irregular surface; between this and the connective tissue is a narrow gap containing two elongated osteoclasts (A and B) and a few red cells. The osteoclasts have been pulled away from the bone by shrinkage of the section; A has a very irregular surface corresponding approximately with the irregularities of the bone surface. Part of a blood-filled thin-walled vessel (V) is seen in the fatty marrow (*top right*). (Paraffin section; haematoxylin and eosin, ×315.)

Re-examination of the slides (1957)—As the patient had now been under observation for over ten years and Gorham and Stout's (1955) most useful paper on the histology of the condition had appeared, the sections were further studied.

Gorham and Stout found in sections obtained from eight of twenty-four cases an essentially similar histological picture, the most striking feature being the greatly enlarged thin-walled vessels in the marrow spaces of the affected bone. These were mostly distended with red cells, but some, particularly those in the connective tissue remaining after absorption of the bone, formed an anastomosing system of empty vascular spaces. Gorham and Stout called the condition " haemangiomatosis," but noted that the process did not resemble the formation of ordinary haemangiomata in bones. The mechanism by which the bony trabeculae were absorbed was not apparent, no osteoclasts being recognised.

The sections from the 1949 biopsy of the present case show the intact hyaline cartilage of the glenoid cavity with a thin layer of supporting cancellous bone which is about six millimetres

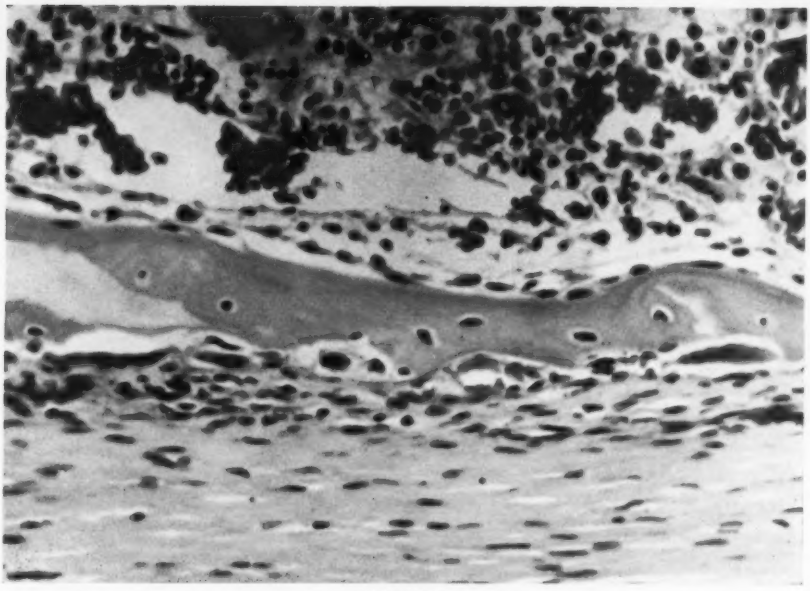

FIG. 10

Normal bone absorption on inner surface of infant's rib, for comparison with Figure 9. Periosteum below; above this a trabecula of bone showing subperiosteal osteoclastic absorption with shallow bays (Howship's lacunae) produced by flattened osteoclasts. A line of these cells (pulled away from the bone by shrinkage) lies between the bone and the periosteum. (Paraffin section; haematoxylin and eosin, ×315.)

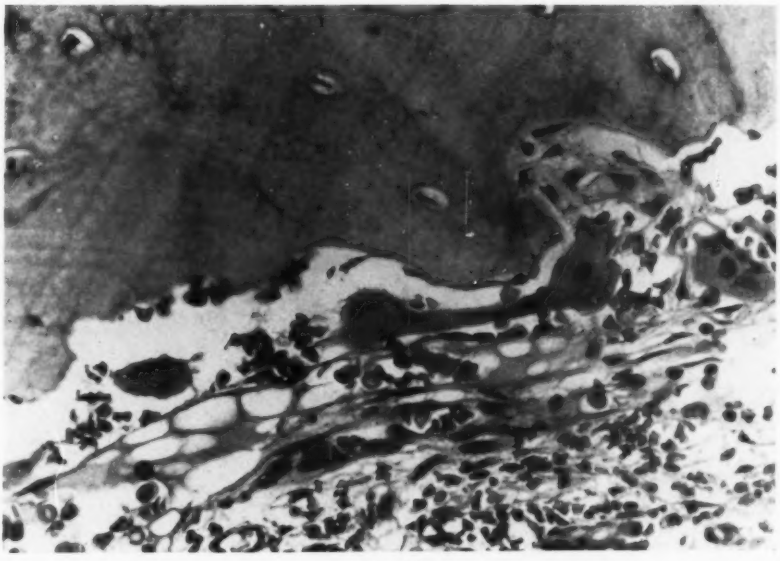

FIG. 11

Another field near that shown in Figure 9; a relatively large dumb-bell-shaped osteoclast and two smaller specimens are seen. The bone surface shows many shallow erosion pits (Howship's lacunae). Below is vascular reticular tissue with some haemorrhage. (Paraffin section; haematoxylin and eosin, ×500.)

thick at one end of the section but tails off at the other end, where only a few minute fragments of bone remain at the base of the cartilage. The spaces in this cancellous bone are filled by fatty marrow containing many relatively large thin-walled vessels distended by red blood cells (Fig. 13). Over most of its free surface (except at one end where the soft tissues have become detached and only a little haemorrhage remains) a thin layer of connective tissue covers the bone, separating it from a little underlying fat and muscle. The trabeculae of the cancellous bone, where they abut on the connective tissue layer, show irregular pitted surfaces, and numbers of osteoclasts, mostly of the flat type, can be seen on, or slightly separated from,

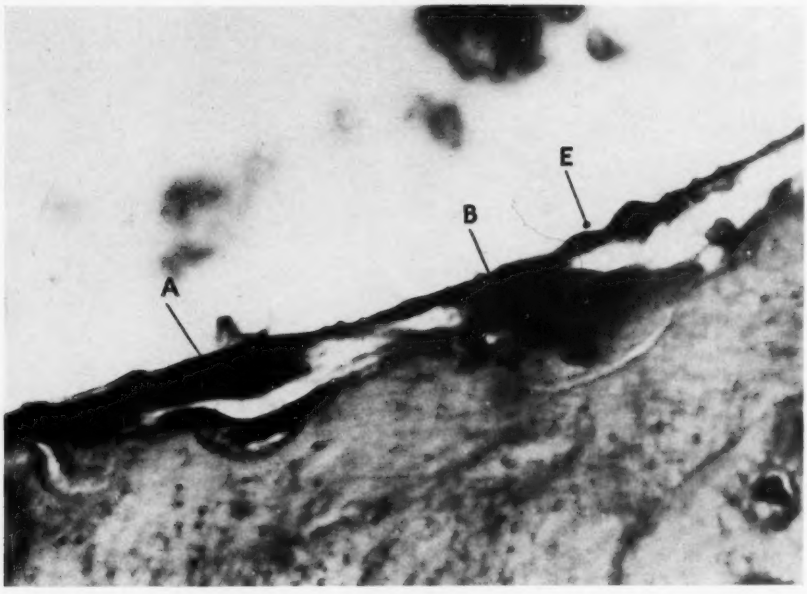

FIG. 12

Osteoclasts lying between wall of a venous sinusoid and the bone surface. Running diagonally across the picture is the endothelial wall of the sinusoid (E); above this is part of its lumen with some scattered clumps of red cells (out of focus). Lying between the endothelium and the bone are two osteoclasts (A and B). The spindle-shaped specimen (A) was inactive at the time of biopsy since it is separated from the bone by three osteoblasts (which are probably starting to fill in the two small pits which they cover). The more rounded osteoclast (B) is active and has produced an erosion pit in which it is partly embedded. (Paraffin section; haematoxylin and eosin, ×950.)

the pitted bone surfaces (Figs. 9, 11 and 12). Numerous distended thin-walled vessels—some containing red cells, others empty—are seen in this connective tissue layer. Neither the underlying muscle nor the fatty tissue on its surface shows signs of increased vascularity. At one point a relatively large very thin-walled vessel or sinus, partly filled with red cells, lies on the surface of the connective tissue layer and in contact with a trabecula, whose adjacent surface shows many shallow pits occupied by osteoclasts, mostly small and flattened, lying between the sinus wall and the bone surface (Fig. 12).

Osteoclasts were found only in relation to those parts of trabeculae that abutted on the connective tissue layer and showed lacunar absorption of their surfaces; none was found on the surfaces of the trabeculae in the vascular fatty marrow, nor did these trabeculae show any lacunar absorption.

Except for a few small trabeculae of imperfectly lamellar bone there was no sign of new bone formation.

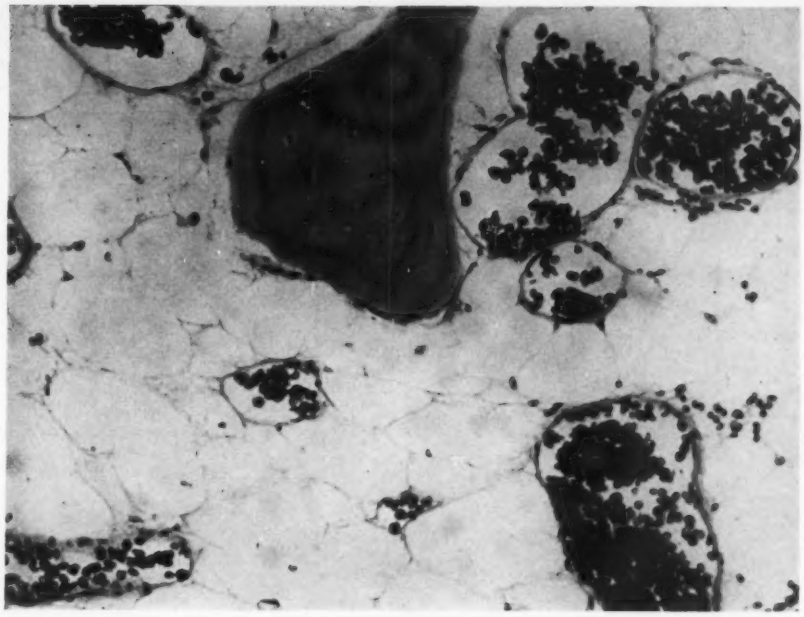

FIG. 13

Enlarged venous capillaries in fatty marrow from cancellous bone in biopsy from this case. (Paraffin section; haematoxylin and eosin, ×200.)

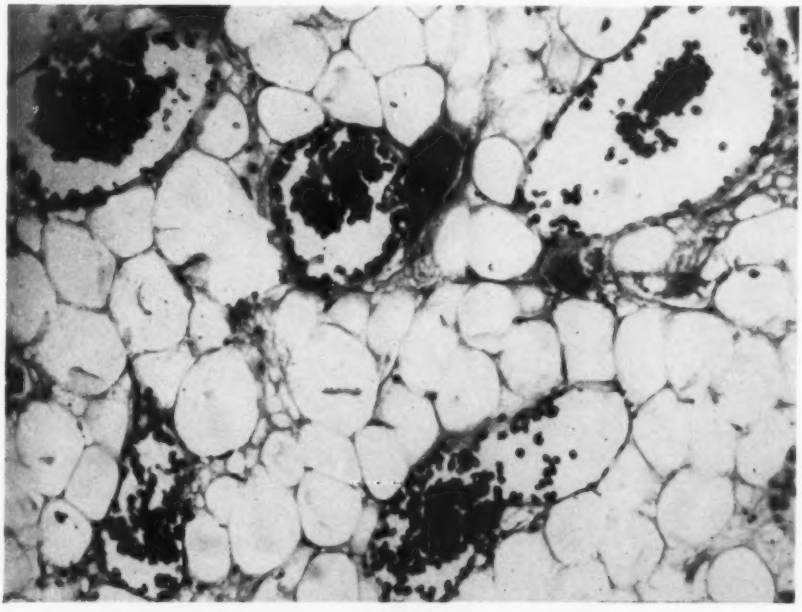

FIG. 14

Biopsy from line of healing four weeks after compression arthrodesis of knee. Venous capillaries similar to those in Figure 13 in fatty marrow of femur near line of healing. (Paraffin section; haematoxylin and eosin, ×200.)

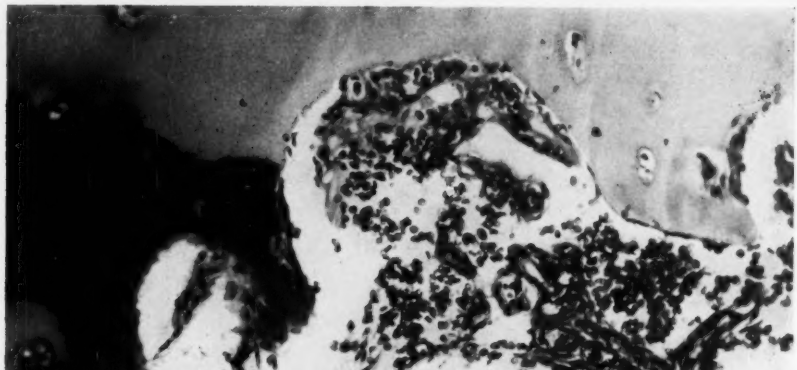

FIG. 15

A small focus of vascular absorption in the base of the articular cartilage of the glenoid cavity. A vascular tuft is seen extending into a bay in the non-calcified hyaline cartilage with its capillary loops in contact with the cartilage surface. To the left is the remains of the zone of calcified cartilage (dark zone). The underlying bone has been absorbed. (Paraffin section; haematoxylin and eosin, × 200.)

FIG. 16

Thin-walled intercommunicating sinusoids (S, S) in the connective tissue at the base of the cartilage at a site where nearly all the bone has been absorbed. The sinusoids contain scattered groups of red cells; smaller vessels extend from them running parallel to the connective tissue bundles on either side. (Paraffin section; haematoxylin and eosin, ×140.)

Where the supporting bone at its base had been largely removed, cartilage was being absorbed at a few points. Figure 15 shows a small vascular tuft which passes through a gap in the calcified zone at the base of the cartilage and is producing absorption of the non-calcified cartilage.

Calcified cartilage, like bone, is commonly absorbed by osteoclasts, but it is generally accepted that non-calcified cartilage can be removed without the development of multinucleated cells—apparently by masses of small cells (possibly endothelial) at the surface of the vascular buds—a process of vascular absorption.

In some places from which the bone at the base of the articular cartilage had disappeared, leaving only connective tissue bundles, the intervening spaces are filled by intercommunicating vascular spaces (" sinusoids "), walled by only a single layer of endothelium. Some contained clumps of red cells; others were almost empty (Fig. 16).

DISCUSSION

Bone absorption—The appearances in our sections correspond in general with those described by Gorham and Stout (1955) but with one important difference: Gorham and Stout found no osteoclasts or any other indication of the mechanism of bone absorption, whereas in our material osteoclasts and Howship's lacunae were present in the deep surface of what remained of the cancellous bone. The appearances here were very like those seen in the normal remodelling of infants' bones, particularly in the areas of active surface absorption such as occur on the pleural surface of the ribs in the metaphysial region (Fig. 10); there is a continuous subperiosteal osteoclastic absorption on the inner (concave) surface while new periosteal bone is being deposited on the outer (convex) surface of the rib.

The pathological absorption in our material appeared rather more active than the normal surface absorption at the most active sites in the ribs of infants a few weeks old, as witness 1) more numerous osteoclasts (ten osteoclasts on a 1·5 millimetre length of bone surface against five on the same length of normal infant's bone) and 2) on the average rather larger osteoclasts (Fig. 11).

There is (Kölliker 1873) a correlation between the size of the osteoclasts and the rate of bone absorption. Where this is rapid the osteoclasts are large and produce deep pits (Howship's lacunae); where it is slow, small flattened osteoclasts produce much less conspicuous shallow pits. Normal osteoclasis in post-natal life is slow as compared with most forms of pathological osteoclasis. The rapid osteoclasis in Paget's osteitis fibrosa, for instance, shows numerous very large osteoclasts and correspondingly large erosion pits. In this case of disappearing bones the histology is not that of a rapid pathological absorption, but indicates a rate of absorption not greatly exceeding that in the normal remodelling process in young children. The synonym " acute spontaneous absorption of bone " is not really appropriate, because in most cases it takes years to absorb a limb bone—in this case approximately eight years to absorb the humerus. This rate of absorption could be compensated for by a moderate grade of osteoplastic activity, but there is little sign of any active new bone formation either histologically or radiographically—a point noted in Gorham and Stout's series of cases. The very poor osteoplastic activity in the affected bone seems to be an important part of the disease, and would account for the gradual removal of the affected bone with a persistent osteoclasis of only moderate activity. (As the condition advanced and the limb was put out of action, disuse also must have played a part in the atrophy.)

As already noted, in our slides osteoclastic absorption was confined to the exposed surface of the cancellous bone fragment, no osteoclasts or Howship's lacunae being found on trabeculae away from this surface.

Vascular changes—The large, thin-walled, distended vessels resembling those described and figured by Gorham and Stout (1955) were conspicuous in the fatty marrow of the cancellous bone in our biopsy material. These distended vessels, though much larger and more conspicuous,

were not more numerous than the small capillaries in samples of normal fatty connective tissue and fatty bone marrow. The fatty connective tissue just outside the bone in our case (distant only a few millimetres from the congested bone marrow) showed only the usual fine capillaries, carrying as a rule only a single line of red cells. Counts of these capillaries showed that, though inconspicuous, they were more numerous than the distended vessels in the bone marrow. Enlargement of the normal capillaries would, therefore, be sufficient to produce the appearances seen in the marrow spaces.

The vessels that enlarge in this way are the venous capillaries which, though normally inconspicuous in fatty marrow, can increase in size under various conditions to form large thin-walled "sinusoids." Various types of cellular activity in the marrow will produce such an enlargement, which is a normal event when fatty marrow is replaced by the much more active haemopoietic marrow whose sinusoids have been studied by various workers (Maximow 1927). A simple reaction in bone is sufficient to enlarge the venous capillaries in the neighbouring fatty marrow to a degree comparable with that seen in these cases. One of us (S. L. B.) has studied this point in a series of biopsy specimens taken from the site of bony union four weeks after the operation of compression arthrodesis (Charnley and Baker 1952). Here there is a rapid aseptic healing limited to a narrow zone of repair tissue (2–3 millimetres wide in the specimen from which Figure 14 was taken). Figure 14 shows the neighbouring fatty tissue with venous capillaries as large and as numerous as those in our case of disappearing bone shown in Figure 13. In the region from which Figure 13 was taken the diameters of the vessels ranged from 65 to 105μ; in that from which Figure 14 was taken the range was 84 to 126μ; and in Figures 2-D, 7 and 9 in Gorham and Stout's paper the range was 75 to 100μ. The only difference noticeable between the vessels seen in Figure 13 and those in Figure 14 is the slightly thicker walls in the former. (This slight difference is probably real but differences in section thickness can produce such an effect.)

At some sites from which the bone had disappeared Gorham and Stout described and illustrated what appeared to be a network of intercommunicating empty vascular spaces lying between the collagen bundles. In Gorham and Stout's Figures 3-C and 6-C, which showed the appearance best, the bundles were for the most part cut transversely. Our Figure 16 shows what appear to be the same spaces with the collagen bundles cut mainly longitudinally. Thin-walled spaces cannot, of course, exist empty in the body, and they must have been distended with some fluid, presumably venous blood which filled the waste space remaining after removal of the bone. The process would be a gradual one, and the venous capillaries or sinusoids would enlarge to fill the vacant space.

Comments—The vascular changes in our case and in most of those discussed by Gorham and Stout have not the features of a neoplastic process. Though the term "haemangiomatosis" implies a neoplastic proliferation of blood vessels, Gorham and Stout evidently did not regard the vascular changes in most of these cases as neoplastic. The occurrence of these enlarged blood-filled venous capillaries is a non-specific reaction accompanying various kinds of cellular activity in or near bone.

In our case cellular activity with osteoclasis was present on the eroded surface of the remains of the bone. Gorham and Stout (1955) noted in their discussion that "no osteoclasts can be recognised," but in an earlier publication (Gorham, Wright, Shultz and Maxon 1954) the description of the rib biopsy of their first case stated that "histologically much of the cortical and cancellous bone had disappeared and that which remained was surrounded by osteoclasts." It seems highly probable that osteoclasis was the mechanism of bone removal in all the cases and that Gorham and Stout's material would have shown this if their sections had included surfaces where bone removal was in progress at the time of taking the specimen.

Even if it is assumed that the immediate mechanism of bone absorption in these cases was the usual process of osteoclastic erosion found in the removal of normal and pathological bone, there remains to be determined what upsets the normal balance between absorption

and deposition in the affected region of the skeleton in these cases. Failure of deposition seems an important factor, but the biopsy of our case shows also an increased osteoclasis. Gorham and Stout suggested that the vascular changes are primary and are the cause of the bone absorption. This may be so; but, since increased vascularity is always found with increased cellular activity, both osteoplastic and osteoclastic, the separation of cause and effect seems almost impossible.

As already noted, there was in our biopsy specimen no increased vascularity in the fatty and muscular tissue within a few millimetres of the very vascular bone marrow. This suggests that there was no general vascular congestion (active or passive) of the non-osseous tissues in the affected region.

SUMMARY

A case is reported of a woman in whom the bones of one upper limb have been progressively disappearing during at least eleven years. Investigations have thrown no light on etiology or treatment.

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A CASE OF "MASSIVE OSTEOLYSIS" OF THE FEMUR

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Very rarely part of the skeleton fades away for no apparent reason. Gorham and Stout (1955) named this condition " massive osteolysis " and reported two cases, and they collected twenty-two further cases from the literature.

In all, the outstanding histological feature has been an increase in the number and size of capillaries engorged with red blood cells. The term " haemangiomatosis " was suggested. The process can start in any bone. It begins as a localised erosion, and from this, slow centrifugal absorption of a large piece of the skeleton takes place. Eventually the process becomes arrested, but only after widespread destruction has occurred. The condition seems to be rare enough to justify the report of another case.

CASE REPORT

A girl of eighteen was first seen in October 1951, complaining of pain in the left thigh of gradual onset and six months' duration. The pain was not severe; it varied considerably, but

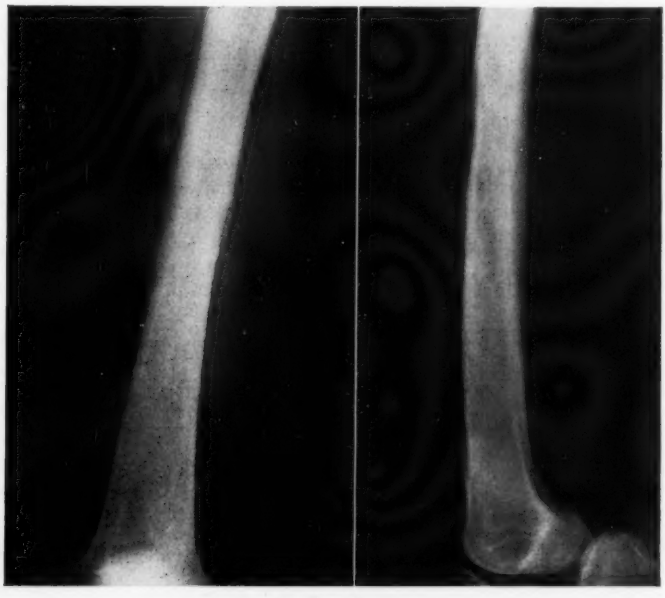

FIG. 1
Initial radiograph, showing early mid-femoral subperiosteal erosion.

was slowly becoming worse. Examination showed an inch of wasting of the left thigh and some deep tenderness at the junction of the middle and lowest thirds, with an impression of local thickening. Radiographs showed subperiosteal erosion of the femur (Fig. 1).

FIG. 2

FIG. 3

Figure 2—Pathological fracture through site of biopsy; further erosion lower in the femoral shaft can be seen beginning. Figure 3—Sixteen months later the fracture has shown signs of union but further extensive erosion of the femoral shaft has caused collapse.

FIG. 4

FIG. 5

Figure 4—Seventeen months after excision of the eroded area, showing incorporation of the graft. Figure 5—Radiograph immediately before the final operation, showing fresh erosion taking place in the femur, both above and below the graft.

Biopsy was undertaken. The periosteum, though slightly thickened, stripped easily. The bone seemed rough and its cortex thin.

Histology (Dr R. J. R. Cureton)—Sectons were taken of the periosteum, muscle, bone and bone marrow. These showed a large number of prominent vessels, including arterioles, capillaries and vessels whose features were like those of an arteriovenous aneurysm. There was no question of any malignant change.

Progress—A pathological fracture occurred at the site of the biopsy (Fig. 2). This eventually showed signs of union, but, at the same time, erosion progressed to involve a large part of the mid-femoral shaft (Fig. 3). Amputation was offered, but declined.

In May 1953 (nineteen months after the biopsy) the eroded segment of bone was excised and the gap was bridged by a massive autogenous tibial graft. The graft slowly became

FIG. 6

FIG. 7

Figure 6—Macroscopic appearance of the upper end of the femur. Figure 7—Slab radiograph of the lower end of the femur, showing erosion where the graft joins the bone, and also in the graft itself.

incorporated (Fig. 4), and after a further nineteen months the hip spica was removed and a weight-relieving caliper was fitted. This was discarded in July 1956. Two months later the patient slipped and sustained a fracture at the upper end of the graft. This fracture again slowly united, but further radiographs showed erosion taking place at both the upper and lower ends of the grafted area (Fig. 5). Finally, it was decided that the only real prospect of arresting the disease would be by total ablation of the femur.

Operation (May 1957)—Skin flaps were cut as for amputation through the thigh at the site of election, and the incision was extended upwards on the outer side of the thigh, finally splitting the gluteus maximus. The whole femur was then removed extraperiosteally (Seddon and Scales 1949) and the upper two-thirds replaced by an implant of cobalt-chrome-molybdenum

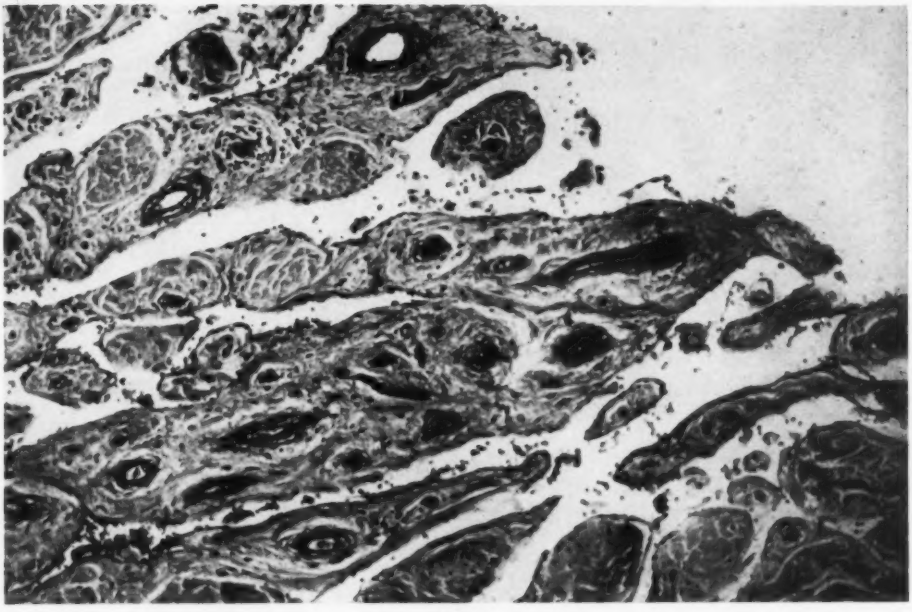

FIG. 8
Photomicrograph of part of the wall of the femoral lesion, showing a network of vascular fibrous tissue. (×95.)

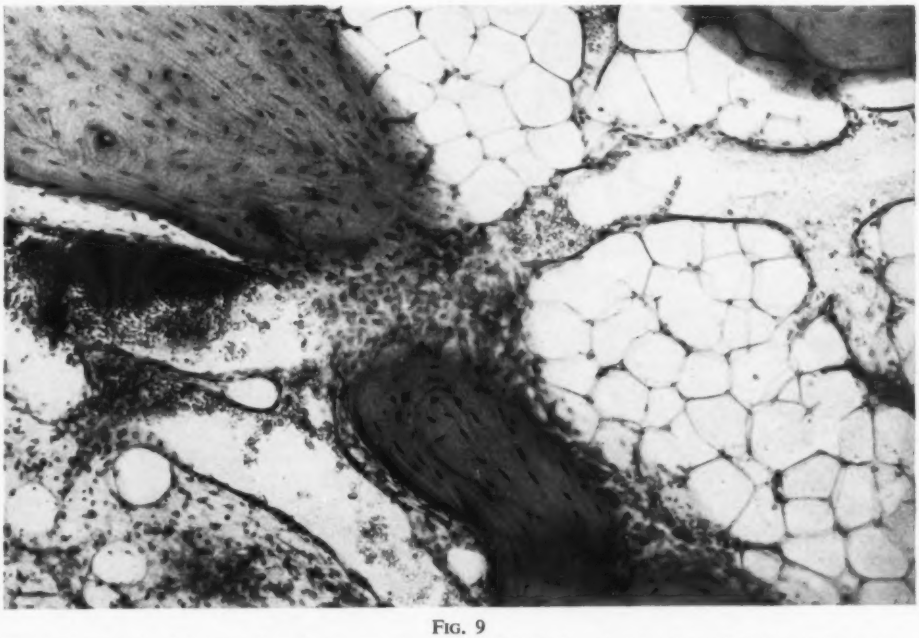

FIG. 9
Photomicrograph of the cancellous bone near the margin of the lesion, showing large dilated vascular spaces. (×95.)

alloy to which the muscles were attached so that the patient could wear an above-knee artificial limb. She was fitted with this six months later.

The macroscopic appearance of the upper end of the femur is shown in Figure 6.

Histology—Dr H. A. Sissons reported that at the margins of the graft the upper and lower parts of the femur showed cystic spaces occupied and lined by cavernous or sinusoidal tissue (Fig. 6). This tissue appeared to be eroding the adjacent bone and, in the region of the femoral neck, showed some extension into the neighbouring soft tissue. The graft itself showed erosion by the vascular tissue. Some patches of acellular necrotic bone were present in it, but the bulk was replaced by newly formed cellular bone (Fig. 7). The characteristic vascular tissue present in the lesion of the femur suggested that it should be regarded as a type of angioma of bone (Figs. 8 and 9).

Most grateful thanks are due to Mr H. Jackson Burrows for permission to report this case, and for all the helpful advice and criticism he has given in the preparation of this report; to Dr R. J. R. Cureton and Dr H. A. Sissons for the histological details and to the Department of Medical Photography of the Institute of Orthopaedics for the illustrations.

The prosthesis, designed by Dr J. T. Scales of the Department of Biomechanics and Surgical Materials of the Institute of Orthopaedics, was cast in " Vinertia " by Messrs Trucast Ltd. and machined by Messrs Deloro Stellite.

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NOTES ON A RARE CASE OF ESSENTIAL OSTEOLYSIS

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In 1944 a schoolboy, then aged thirteen, was wounded slightly in the right foot by a nail, which caused a moderate lymphangitis of the leg. A month before, while running, he had felt his right leg give way momentarily. A few days after the injury to his foot, while he was bathing in the sea and already recovered from the trauma, he suffered a sudden pain in the right groin which increased rapidly and became very severe a few hours later. There was marked pyrexia, with rigors, and examination showed a diffuse tender swelling of the right thigh and limitation of hip movements. Under sulphonamide treatment the fever and pain subsided and full hip movement was regained within a week. However, from that time, whenever the patient walked his leg would become affected with oedema of the thigh and knee, which would disappear with rest.

A few days later the same clinical picture of pyrexia preceded by rigors recurred; hip and knee movements were greatly impaired, and there was lymphadenitis in the groin. Sulphonamide treatment was resumed and the patient felt well after three days, with disappearance of the

FIG. 1
Photograph showing the marked shortening of the right thigh.

fever, but rest in bed was continued for two weeks on account of the persistent inguinal lymphadenitis. Painless, mobile lymph nodes could be felt, the size of small cherries, without any cutaneous reaction.

The patient returned to his normal activities, but he continued to note a swelling of the right thigh, spreading downwards to the knee, especially at the end of the day. This oedema gradually became more persistent and severe, until it continued to exist even with rest.

FIG. 2

FIG. 3

Figure 2—Right thigh showing total destruction of the upper two-thirds of the femur. Figure 3—Pelvis (1950) showing disappearance of a large part of the right half of the pelvis and sacrum, with beginning destruction of the left pubis and ischium.

About four months later a yellowish discoloration appeared on the inner aspect of the thigh, changing to a purplish tone before disappearing some time later. The area was hard but painless. Although the thigh and knee remained swollen, the patient could walk without limping, could run well and had no pyrexia. Radiographs did not reveal any local abnormality.

FIG. 4
Right knee and upper parts of tibia and fibula in 1950. Partial atrophy of fibula.

Despite massage, anti-streptococcal vaccines and lumbar sympathetic infiltrations of procaine, the clinical picture continued to progress, with gradually increasing swelling. Function was but little impaired until August 1945—eleven months after the onset—when a spontaneous fracture occurred at the junction of the middle and uppermost thirds of the right femur. Radiographs showed marked rarefaction of the femur and a complete oblique fracture. A tentative diagnosis of Ewing's sarcoma was made. A plaster was applied and radiotherapy given in two courses with an eight months' interval. The patient's general health remained good, but radiographs showed progressive rarefaction of the femur. The lymph nodes in the groin could no longer be felt.

In April 1947, twenty months after the fracture, hormone treatment with hexoestrol was begun and maintained for about a year. This caused a marked gain in weight, with considerable gynaecomastia.

Successive radiographic examinations revealed a spreading of the bone destruction downwards to involve the knee joint, and upwards to involve the pelvis, including the sacrum.

FIG. 5

FIG. 6

Figure 5—Right knee and upper parts of tibia and fibula in 1956 showing increased destruction of fibula. Figure 6—Pelvis in 1956 showing further destruction of the right ilium and left pubis and ischium.

At this time the plaster had to be discarded because of intolerance of the skin. The limb was very much shortened, and presented grossly abnormal mobility.

In January 1950, about six years after the onset of the illness, the patient first came under our care. He was then nineteen years old and very fat. He moved about in a wheel-chair, resting on what remained of his pelvis, with his right leg in a splint. Examination revealed nothing except the abnormality of the right lower limb, which, deprived of all voluntary movement, was greatly deformed and about twenty centimetres shorter than the left (Fig. 1). The skin, covered with hair and without any trophic changes, was wrinkled with transverse folds, and was strangely mobile over the deep layers. The muscles were flaccid. The right foot, though smaller than the left, was not deformed and could be flexed and extended. The whole limb was painless to the touch and to pressure, and the consistency of the tibia and patella was normal, although the latter was deformed. The knee was totally ankylosed. The limb was strikingly flexible and could be bent painlessly in all directions, the movements being limited only by the soft parts.

Radiographic examination—Radiographs (1950) showed that the upper two-thirds of the right femur, together with the lower half of the right innominate bone and part of the sacrum, had completely disappeared (Figs. 2 and 3). The destructive process had extended across the symphysis pubis to the left half of the pelvis. The right ilium, the lowest third of the right femur, and the right tibia and fibula were markedly rarefied (Fig. 4). A skeletal survey showed no alteration of any other bone (Fig. 7). Pyelography did not reveal any abnormality of the renal tract. Later radiographs, taken in 1956 and showing increased destruction of bone, are shown in Figures 5 and 6.

Investigations—Repeated laboratory tests, including serum calcium (10·4 milligrams per cent), serum phosphorus (3·1 milligrams per cent) and alkaline phosphatase (4·2 Bodansky units) estimations, blood counts, erythrocyte sedimentation rate, urine examinations, Sulkowitch's calcium excretion test, Wassermann reaction, and blood sugar and blood urea determinations, were all normal. The basal metabolic rate was plus 21 per cent. The electrophoretic curve, for a total value of 6·28 grains of protein, was within normal limits. Test meal was normal, as was the cerebrospinal fluid. Calcium balance studies showed that there was no abnormal loss of calcium from the body. The only finding that was constantly abnormal was the blood magnesium concentration, which was low at 1·1 milligrams per cent.

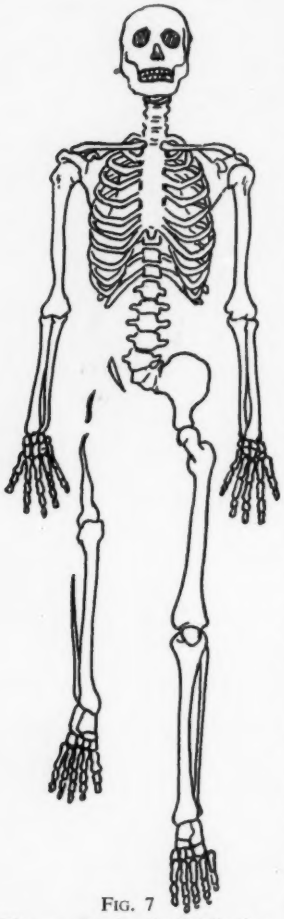

FIG. 7
Diagram showing the distribution and extent of the bone destruction in 1956.

Biopsy—During the past six years we have made three biopsies, the first specimen being removed from the crest of the right ilium, and the last two from the lowest third of the femur.

The most recent biopsy, in March 1957, included fragments of the femur, of muscle and of nerve tissue. The exposed region appeared normally vascularised and the remains of the right femur (little more than the lowest third) were approached through poorly developed muscular masses. The appearance and consistency of the bone were normal and the upper extremity of the bone terminated sharply, like a pencil point.

Histological studies (Professor J. da Silva Horta) (Figs. 8 to 11)—The remaining bone contained Haversian canals that were widened and filled with fatty marrow. The lamellae were for the most part regularly arranged, but there were cement lines that were sometimes numerous and oriented in different directions, resulting in irregular mosaic figures. At the edge of the bone, adjacent to the fibrous type of tissue to be mentioned later, there was newly formed lamellar bone, but the lamellae were disposed at right angles to those of the old bone substance. This feature was seen in all the sections, regardless of the method of preparation, but was seen especially clearly in silver impregnations (Tibor-Pap). In the new bone osteocytes were numerous. There was also osteoid substance at the edge of the bone, but only in small quantity. The bone substance presented an intense rearrangement in which the process of destruction exceeded that of repair. In addition to the widening of the Haversian canals, the initially compact bone was transformed into spongy bone; the intervention of osteoclasts was practically nil. The essential process was a replacement of bone substance by an unusual connective tissue. This was a basophilic substance containing connective fibres with an undulated, winding trajectory. Some of these fibres were thick and slightly eosinophilic, staining blue and yellowish grey by Van Gieson's stain. Many of these fine and coarse fibres were impregnated by silver, and there were elastic fibres among them.

Isolated masses of this peculiar fibrous tissue appeared in the midst of ordinary loose connective tissue, just as if they represented already demineralised trabeculae of bone, but with a perfectly preserved contour which permitted their individualisation. The limits of one of these masses were marked by curved fibres, just as in a capsule. These masses of special tissue contained a few scattered capillaries and a few cells, many with pyknotic nuclei. There

FIG. 8

Low power photomicrograph of a section from the right thigh showing an intense osteolytic process. The bone trabeculae are transformed into a matrix of unusual appearance, in the midst of which may be seen the remains of mineralised trabeculae (arrows). (Haematoxylin and eosin.)

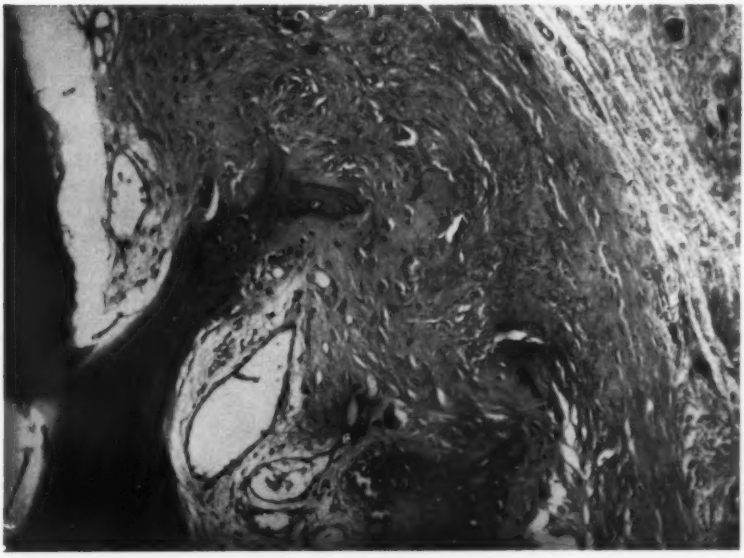

FIG. 9

Another part of the material, showing the same osteolytic process. The same special matrix is seen, already decalcified, with bony trabeculae in the course of transformation. (Haematoxylin and eosin.)

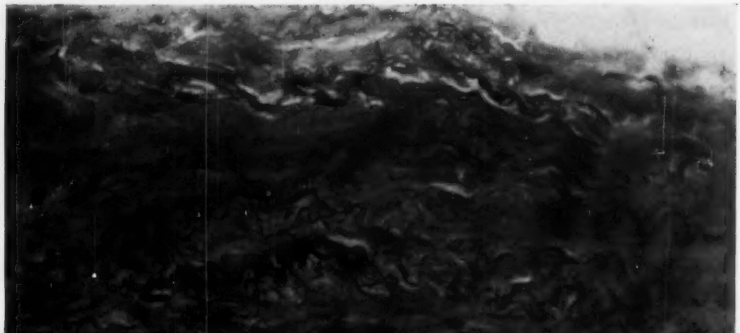

FIG. 10
The matrix referred to in the preceding figures contains undulated fasciculi. (Haematoxylin and eosin; medium magnification.)

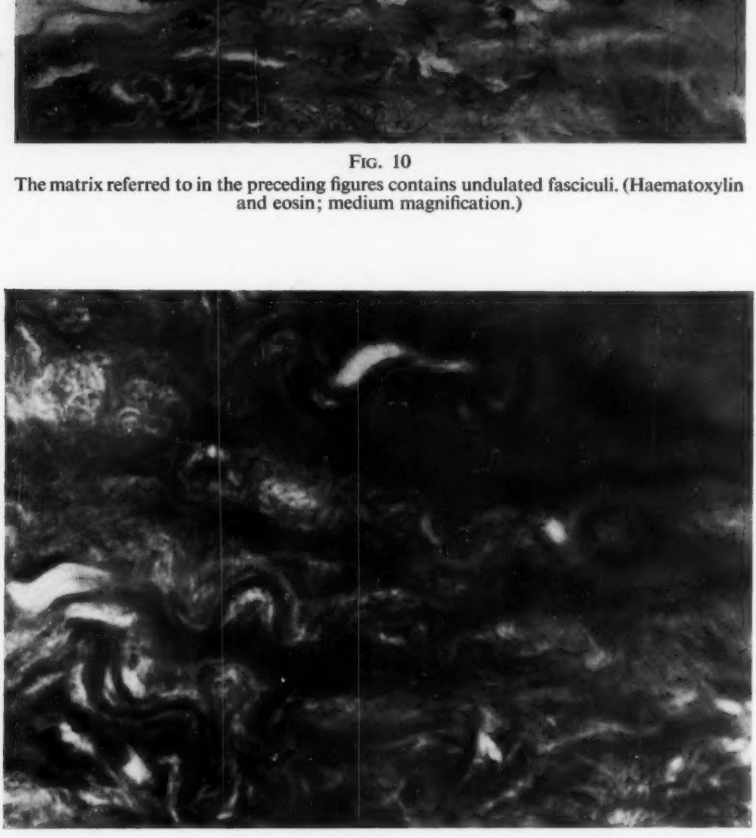

FIG. 11
The same undulated and tangled fasciculi as those seen in Figure 10. (Mallory stain; high magnification.)

were interstitial spaces in this tissue which communicated with large empty areas. Sometimes small fragments of the tissue described were scattered throughout a loose oedematous connective tissue. In other places the bone was continued either by dense fibrous connective tissue, with a variable number of histiocytes and fibroblasts, or by slightly irregular cartilaginous tissue, or by a special tissue that was in general fibrous, but which contained one or another element of the type of cartilaginous cells. With the Mallory or Tiber-Pap stains it was verified that at certain points the fibres of the bone substance were continued by those of the dense or cartilaginous fibrous tissue, and also by those of the fibrous basophile tissue. There was no evidence of active osteoclasis; one or two osteoclasts might be found in the fibrous eosinophil tissue and in a very restricted zone close to small remains of bony trabeculae that persisted in the fibrous mass. There was active bone marrow in several places.

The striped muscle presented important alterations: some fibres showed a variable degree of atrophy, others showed loss of striation, and still others were transformed into intensely eosinophil hyaline masses. There was slight proliferation of the nuclei of the sarcolemma, and infiltration around the vessels by small round lymphoid cells. There was some proliferation ex-vacuo of the adipose and fibrous tissues. The latter possessed slightly undulated fibres, but they were basophilic.

In the loose connective tissue surrounding the muscle, and separating it from blood vessels, there were several lymphocytic infiltrations. In this same tissue there were irregular spaces filled with fibrinous detritus which was either free or disposed in fasciculi along the edges of the spaces.

The nerves were deeply infiltrated by a basophilic metachromatic (toluidine blue) substance. The axons were intensely altered, with disappearance of various segments, pronounced swellings, sometimes the beginning of rosary formation—that is, with widened portions alternating with very fine ones. In addition to this, there were several isolated segments of the axons which were curved in an arc. The myelin sheaths were relatively intact.

TABLE I

COMPARATIVE ANALYSES OF FLUID FROM THE PATIENT'S THIGH AND BLOOD SERUM

Constituent	Content (*milligrams per cent*)	
	Aspirated fluid	Blood serum
Calcium . . .	8·7	10·1
Phosphorus . .	3·1	2·7
Alkaline phosphatase	3·1	2·0

Further treatment and progress—On three separate occasions we withdrew, from the depths of the thigh and of the sacral region, sero-sanguineous fluid in abundance. The study of this fluid showed that most of the cellular elements were red blood cells, with lymphocytes and polymorphonuclear cells also present, with isolated macrophages and cells of the histiocyte type. The electrophoretic curves of proteins obtained from blood serum and from this fluid were identical. Chemical analysis of the fluid supported the probability that the fluid was derived from the blood (Table I).

DISCUSSION

With no precise knowledge of the etiology or pathogenesis of the condition, the anatomical and histological observations can help us but little to explain its nature.

In the presence of such results, some investigators have suggested an active hyperaemia conditioning the reabsorption of the bone and its substitution by fibrous tissue. We meet expressions such as: " fibrous tissue rich in blood vessels," " marrow replaced by fibrous tissue rich in vessels," " appearance of fibrocystic disease," in the reports of the rare cases that have been examined from the histological point of view.

In the opinion of Gorham and Stout (1955) the active vascularisation would be responsible for the disturbance of the osteoblast-osteoclast balance, with predominance of the destructive activity in relation to the laying down of bone, resulting in bone resorption. In our case the part played by osteoclasts was practically nil, as judged from the three pathological specimens. In some cases the disease seems to be associated with a haemangioma. Leriche (1937) suggested that the process might take place in the presence of hyperaemia caused by injury or infection, and that the increase in local arterial circulation might be the cause of the osteolysis. But, we ask, might not the hyperaemia, if present, be the consequence of the process, a secondary phenomenon? In the three biopsies made in our case, we did not observe any sign of local increase in vascularity.

Mouchet (1943) affirmed that injury, which has been a feature in some cases, is always slight, has no effect on the genesis of the condition and has no greater significance than that of revealing a disease already in evolution.

Leger, Ducroquet and Leger (1949), Coste and Gaucher (1943) and others have suggested that the condition might be of neurogenic origin, associated with or depending upon a lesion of the central nervous system, of the medullary autonomic centres or of the hypothalamic centres.

In our case it seemed as if the stroma, the osteoid tissue, had lost the capacity to fix electrolytes and had liquefied its muco-protein structure by an enzymatic process.

In most of the twenty-five cases of this obscure disease that have been reported the patients have been children or young adults. The sites at which the disease has started include the clavicle, sternum, ribs, humerus, radius, ulna, small bones of the wrist, metacarpal bones, fingers, maxilla and mandible, bones of the foot, femur and pelvis. In our case the lysis involves the fibula, the femur, the ilium and the sacrum on the right side, and has already destroyed a large part of the ischium and the pubis on the left. The upper part of the right tibia and the lower lumbar vertebrae are already seriously rarefied.

The evolution of the disease is, as in our case, lengthy, painless, afebrile and often complicated by fractures.

The breakdown of the bone resists all the therapeutic measures tried so far, and tends to progress (Figs. 5 and 6).

In this case we started with androgens, continuing their administration for a long time, up to a total of 2 grains of testosterone propionate, as a result of which the patient lost some 20 kilograms of weight. Later we added calcium fluoride, without any visible benefit. We also tried suprarenal extracts (in view of the importance of the function of these glands in the elimination of electrolytes), vitamin D_2, aluminium acetate solution, ultra-violet radiation and ionised calcium, and somatotrophic hormone. The prolonged administration of placental extracts, of vitamin B_{12} and of other vitamins and amino acids, was useless. We made transfusions of placental blood and a series of transfusions of blood obtained from young individuals whose growth had not yet terminated, with the idea that the blood of such individuals would be richer in bone factors than that of mature individuals. These transfusions were of no avail. Since experimental fractures have been found to heal more rapidly when the animal receives small doses of Bogomoletz serum (Straus *et al.* 1946) we tried this serum for a long time, but without any benefit.

We did not attempt a bone-grafting operation, fearing that the graft would only disappear, as described by Dupas, Badelon and Dayde (1939).

SUMMARY

1. A case of essential osteolysis, progressing for thirteen years and involving the right femur and pelvis of a boy who was thirteen years old at the time of onset, is presented.
2. The histological appearance of fragments of bone and soft tissue removed from the affected thigh is described.
3. No evidence that might throw light on the etiology of the disease was obtained from biochemical investigations or from the histological studies.
4. None of the many lines of treatment that were tried had any effect upon the progress of the disease.

A clinical report on this patient was published simultaneously by Dr João José Ferro and by Dr Barros do Amaral in 1950. We consider that this further report on the same patient is justified by the rarity of the condition and by the further biochemical and histological investigations that we have carried out.

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THE FLEXOR-EXTENSOR TRANSPLANT OPERATION FOR CLAW TOES

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Clawing of the toes, like that of the fingers, is the result that one would predict from paralysis of the intrinsic muscles. Nevertheless it has not been possible in most cases of claw toes to demonstrate any actual paralysis or atrophy of these muscles (Taylor 1951).

Strong action of the interossei and lumbricals was considered to be of the highest importance in the function of the foot by both Lambrinudi (1938) and Girdlestone (1947), each of whom devised an operation to restore or replace this action when, as in the case of claw toes, it is seemingly lost. In both operations the power of the long flexors, which in claw toes becomes a deforming force, is put to work to perform the function of the "lost" intrinsics.

In the Lambrinudi operation, by arthrodesis of the interphalangeal joints, the long flexors are persuaded to exert all their action on the metatarso-phalangeal joints. The Lambrinudi procedure is too often regarded as a mere "toe-straightening operation"; but far from simply correcting a static deformity it produces a dynamic redistribution of power which restores function as well as improving appearance. Nevertheless, to fuse healthy joints in the absence of true paralysis is a step that does not commend itself to everybody, and for that reason the Girdlestone operation may appear theoretically more acceptable. There is, however, considerable difference of opinion about the practical merits of the operation, and this uncertainty has prompted the present study of the results of the flexor-extensor transplant.

It would be most instructive to compare the results from the two types of operation but this is not possible unless comparable cases are treated. Here, as elsewhere, it has been customary to confine the tendon transplant operation to cases of mobile clawing of the toes and to perform multiple arthrodeses only when the toes are rigidly clawed. This paper therefore does not purport to discuss the relative merits of the two operations, but merely to review the results of the Girdlestone procedure in those patients in whom it appeared to be the treatment of choice.

MATERIAL

In the records of the Orthopaedic Department of the Royal Victoria Hospital, Belfast, for the ten years 1947–56, information was available on thirty-five patients subjected to the Girdlestone flexor-extensor transplant. Twenty-three of these patients attended for review, and since seventeen had had both feet operated upon there were forty feet in which the result could be assessed subjectively and objectively. The records of the twelve patients who could not be traced were not detailed enough to permit reliable assessment except in three cases. The objective assessment previously made in these three cases (two bilateral, one unilateral) brought the total of feet assessed objectively to forty-five.

Of the total number of twenty-six patients thus included in the study, thirteen were male and thirteen female. Ages ranged from thirteen to fifty-five years, but most of the patients were in their late teens or early twenties.

METHOD

The operation was carried out as described in detail by Taylor (*loc. cit.*) with only two minor variations: 1) division of the extensor tendons and the posterior capsule of the metatarso-phalangeal joints was carried out as a part of the set operation and not as a preliminary procedure; and 2) the Lambrinudi splint was not used because of the danger of sepsis and ischaemic complications from the necessary stay sutures. It was found that the

toes could be equally well controlled with an extended plaster applied with strong upward pressure beneath the metatarsal heads and with the toes held in flexion at the metatarso-phalangeal joints (Fig. 1).

Each patient was questioned about his complaints before the operation and about the nature and degree of his residual symptoms, if any, at the time of review. Each patient was also asked to give his general assessment of the result of operation. So that these "subjective

FIG. 1

To show the type of plaster used after operation. (The right foot illustrates the degree of clawing present before operation.)

assessments" might be classified the patients were shown a card (Table I) from which to choose one of five alternative descriptions of the result.

In the objective assessment the following factors were considered: 1) the presence of symptoms, their type and degree; 2) the shape of the toes; 3) the mobility of the toes; 4) the functional efficacy of the transplant as judged from the presence, range and power of active plantarflexion at the metatarso-phalangeal joints; 5) the presence or absence of callosities over the proximal interphalangeal joints and under the metatarsal heads; and 6) other phenomena present only in certain cases.

On these criteria, the result was assessed as excellent, good, fair, poor or bad.

RESULTS

On the whole the patients had a somewhat better opinion of the operation than had the examiner. Table I shows the proportion of feet in each category of result as determined by the patient. Worthwhile improvement (shown in the first two categories) constituted 60 per cent.

Table II shows the result as determined by the examiner (by personal examination in forty feet, from adequate notes in five). Here the first two categories together accounted for only 51 per cent of the feet. This is perhaps surprising because other abnormalities besides

claw toes were frequently present in the feet operated on (most often pes cavus and hallux valgus; occasionally pes valgus and tight tendo calcaneus) and were, of course, uninfluenced by the operation. It was recognised, however, that persistence of these abnormalities would

TABLE I

PATIENTS' ASSESSMENT OF RESULT (FORTY FEET)

Group	Description of result	Number of feet	Per cent
1	Complete success .	4	10
2	Marked improvement	20	50
3	Slight improvement .	8	20
4	No change . .	8	20
5	Worse than before .	0	0

TABLE II

OBJECTIVE ASSESSMENT OF RESULT (FORTY-FIVE FEET)

Result	Number of feet	Per cent
Excellent .	5	11
Good . .	18	40
Fair . .	8	18
Poor . .	10	22
Bad . .	4	9

tend to lower the patients' estimate of the value of operation, and care was taken to explain that their opinion was required of the result only so far as the forefoot was concerned. Even so, a few patients were less satisfied with the result than an objective appraisal warranted. In particular, a youth with "gorilloid" feet, in whom the objective result in the small toes was

FIG. 2

Case 1—The extensor tendons have regenerated after operation, with consequent recurrence of the deformity.

regarded as excellent, rated the result in category 4 ("no improvement") because of the unaltered appearance of the abducted hallux. In most of the other cases the rating given by the patient corresponded closely to that allotted by the examiner.

FIG. 3
Case 2—Left foot before operation.

FIG. 4
Case 2—Appearance of foot after operation. There is a marked improvement in the shape of the toes, and the callosities over the interphalangeal joints are much less obvious.

One other case deserves special mention: that of a patient with marked neurological disturbance of the legs, associated with congenital abnormalities of the spine. This patient classed the result under the category " no change," and the objective assessment was " bad." The patient subsequently had all the toes of both feet amputated. It seems clear that this case was never a suitable one for operation and might justifiably be excluded from the series.

FIG. 5

Case 3—Photographs showing the range of active extension and flexion of the toes after operation. There is a good range of movement at the metatarso-phalangeal joints, but note the poor movement at the interphalangeal joints.

DISCUSSION

The operation cannot be regarded as universally successful. Three causes of imperfect results demand further consideration.

Metatarsal pain and callosities—In no case in which these had been prominent before operation was there significant improvement. Both the pain and the callosities under the metatarsal heads persisted in only slightly diminished degree, even when the transplant appeared to be functioning well.

Stiffness of the interphalangeal joints—This was a common finding. Although passive movement of the metatarso-phalangeal joints was nearly always free, and active flexion of the metatarso-phalangeal joints was good in over half the cases, the interphalangeal joints were stiff as regards both active and passive movement in 60 per cent, whether or not the shape of the toes was satisfactory. Although this may impair the objective result it may not detract from the functional result, as will be mentioned later.

Regeneration of extensor tendons and recurrence of deformity—This was noted in almost 20 per cent of cases. The tendons seem to reattach themselves in a way that makes it appear as if the operation had never been done. In the case illustrated (Fig. 3) there is no doubt that the extensor tendons and the posterior capsule of the metatarso-phalangeal joints were divided. Strange to say, similar regeneration has not occurred in the opposite foot, which was dealt with by an exactly similar technique, and by the same surgeon. I have been unable to find any satisfactory explanation for this perverse and capricious behaviour of the extensor tendons.

Advantages—Despite the drawbacks that have been mentioned the operation has several advantages.

Shape of the toes—This was often markedly improved (Fig. 4), with consequent easier fitting with comfortable shoes. A frequent complaint before operation, especially among women, was the great difficulty in " getting shoes to fit." To many the solution of this problem alone made the operation well worth while.

Corns—Although metatarsal callosities were generally little relieved by the operation, corns over the interphalangeal joints often disappeared. Even when they persisted they generally became painless.

Walking—This was improved in many instances. The improvement is attributed to the restoration of " intrinsic muscle " action in the toes—that is, active flexion of the metatarso-phalangeal joints (Fig. 5). The improved function of the foot reduces fatigue.

CONCLUSIONS

The forty-five feet here reviewed were operated upon by five different surgeons with only minor variations in technique. Good and poor results were more or less evenly distributed; so the poor results cannot be attributed to any individual errors of technique. Indeed it was not uncommon to find distinct differences in result in the right and left foot of the same patient, both feet having been treated by the same surgeon by exactly the same technique. One can only conclude that the operation is somewhat unpredictable in outcome, and is on the whole far from being an unqualified success.

One conclusion emerged clearly: that the best results were obtained in the patients with the mildest symptoms. Dissatisfaction with the shape of the toes, difficulty in obtaining shoes, and corns on the toes, were all symptoms that could usually be relieved by operation. On the other hand, marked cocking-up of the toes, severe metatarsal discomfort, or painful callosities under the metatarsal heads were but little relieved by operation.

An interesting finding was the stiffness that so often develops in the interphalangeal joints, probably from scarring. This loss of interphalangeal movement, though unforeseen and at first sight undesirable, may not be wholly disadvantageous; indeed it may even contribute to the success of the operation by converting the flexed jointed toe into a rigid lever, thereby enabling the long flexors to act strongly on the metatarso-phalangeal joints as in the Lambrinudi operation. It is thus possible that the successful Girdlestone operation works (at least partly) on the Lambrinudi principle. But this fortuitous stiffness is inferior to that produced by deliberate arthrodesis because the toe may become stiff in flexion. Although a true comparison is not possible here it seems likely that the Lambrinudi operation is in that respect the more satisfactory, and it is the operation of choice when the flexor-extensor transplant operation has failed.

SUMMARY

1. Patients subjected to the flexor-extensor transplant have been reviewed. The results in forty feet were assessed by direct questioning and examination, and the results of a further five operations were assessed from the records.

2. Worthwhile improvement was gained in a little over half the cases. The chances of success are greatest when the symptoms are mildest.

3. The operation appears to be inadequate in certain respects and it seems doubtful whether it has any advantage over the multiple arthrodesis (Lambrinudi) operation.

I wish to thank Mr R. J. W. Withers, Mr N. S. Martin and Mr R. I. Wilson for permission to study their patients and for allowing me the privilege of operating on many of them. I am indebted to Mr R. G. Wood for most of the photographs.

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TREATMENT OF ADDUCTOR PARALYSIS BY HAMSTRING TRANSPOSITION

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Isolated loss of power of the adductor muscles of the thigh can occur as a result of poliomyelitis, but it is much more likely to develop as a complication of an over-enthusiastic adductor tenotomy in the spastic. In an attempt to correct the adduction of the thighs with its attendant severe scissor gait, obturator neurectomy is not infrequently supplemented by adductor tenotomy in the groin. The original deformity may be corrected, but in its place in some cases an even more crippling handicap is left. The previously adducted limbs now lie abducted, laterally rotated and flexed, and thus a child previously capable of assisted walking may be unable to walk at all.

In two such cases I have transferred the hamstring muscles to restore the power of adduction.

CASE REPORTS

Case 1—A man aged twenty-one was admitted with cerebral palsy, spastic in type, complicated by dislocation of the left hip. The patient had already been subjected to many surgical

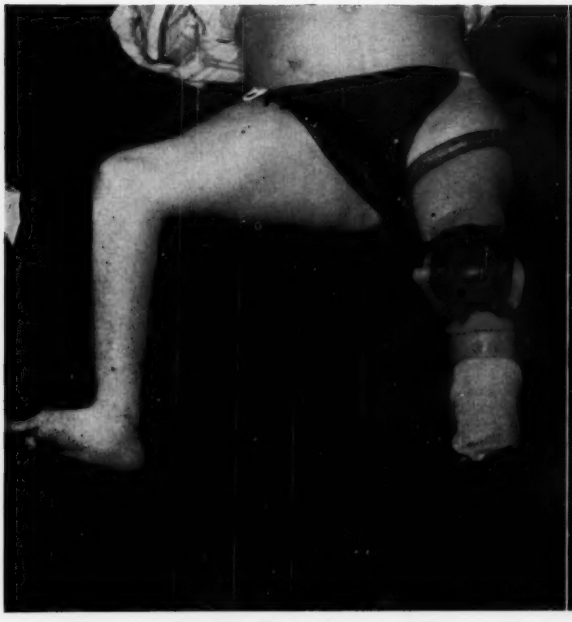

FIG. 1

FIG. 2

Case 1. Figure 1—Before operation. Figure 2—After transfer of the hamstrings to restore power of adduction.

operations in an attempt to correct the dislocation of the hip and to overcome the severe flexion-adduction of both thighs. As a result the left hip had been reduced but, in consequence of a previous extensive adductor tenotomy and obturator neurectomy, the right leg lay in

FIG. 3

Case 2—Photograph showing the marked abduction and lateral rotation deformity on the left side. (Right leg in post-operative plaster.)

FIG. 4

FIG. 5

Case 2. Figure 4—Deformity corrected. Figure 5—Patient walking for the first time in her life.

abduction and lateral rotation (Fig. 1). Treatment by transfer of the right hamstring muscles was done in two stages. Twelve months later he was able to adduct the right thigh to the mid-line (Fig. 2) against gravity.

Case 2—A girl aged twenty-five was admitted with cerebral palsy, affecting all four limbs. In consequence of a previous obturator neurectomy followed by an adductor tenotomy she had lost the power to adduct the thighs. The legs lay abducted, laterally rotated and flexed (Fig. 3).

On each side the hamstring muscles were transplanted in two stages. Six months after the final operation the patient was able to walk in a trolley, and two months later with a walking aid (Figs. 4 and 5).

TECHNIQUE OF OPERATION

The hamstring muscles are suitable for transposition because they possess a comparatively long and mobile neurovascular bundle, because very little realignment is necessary and because their fibre length is comparable to that of the adductor muscles.

FIG. 6

FIG. 7

Figure 6—Hamstring origin mobilised from ischium with a thin slice of bone.
Figure 7—Diagram showing the operation completed.

The tendons of the biceps and of the semitendinosus and semimembranosus are exposed and divided about an inch from their insertions. The semitendinosus and semimembranosus tendons are threaded through a single tunnel drilled on the medial side of the lower end of the femur and sutured to themselves and to the adjacent periosteum or adductor magnus. The tendon of the biceps, which is often broad, may have to be cleared of muscle tissue and split before it can be pulled through a hole drilled on the lateral side of the femur. It is then sutured back to itself or to its divided half and to the periosteum.

Transfer of the hamstring origin from the ischial tuberosity may be performed at the same time, or it may be delayed for two months. This stage is performed most easily with the patient in the lithotomy position. Through a J-shaped incision (the short limb running towards the pubis) the origin of the hamstring muscles is exposed and the neurovascular

bundle is identified to the lateral side. The muscle origin is removed with a thin slice of bone by means of a chisel (Fig. 6). The inferior pubic ramus is identified and the bone exposed subperiosteally as near to the symphysis pubis as possible, care being taken to avoid damage to the urethra or pudendal vessels. The hamstring origin is attached to the symphysis or to the adjacent part of the pubic ramus with a wire suture. This is done best by drilling one or two small holes in the pubis to ensure a firm attachment, which is reinforced further by suturing the reflected periosteum over the hamstring origin with its attached bone flakes (Fig. 7).

After the operation the limb is immobilised in a plaster spica for two months. Thereafter active exercises are begun under the care of the physiotherapist, at first in the pool.

COMMENT

In each of the two cases described a most satisfactory degree of correction of the disabling deformity was obtained. Within a year the limb could be adducted actively to the mid-line; the strength of the transposed muscle was assessed as Grade 3. Both patients were able to walk straight-legged with support—in the second case, for the first time in her life.

I have much pleasure in acknowledging my indebtedness to Professor G. J. Romanes and Dr R. G. Inkster of the Department of Anatomy, University of Edinburgh, for their advice and generous help.

FAMILIAL OSTEOARTHROPATHY OF THE FINGERS

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This paper is concerned with a genetically determined abnormality of the hands characterised by enlargement and loss of mobility of the proximal and distal interphalangeal joints. The abnormality appears in childhood; in later life the clinical and radiographic signs resemble those of some forms of chronic arthritis, but other features are distinctive. The abnormality appears to be one of a group of conditions in which the epiphyses of the bones undergo avascular necrosis, such as Legg-Calvé-Perthes' disease of the capital epiphysis of the femur and Freiberg's disease of the second metatarsal head.

The name of Thiemann is often associated with a rather diverse group of conditions in which there is avascular necrosis of the phalanges of the hands. Usually only one or two of the fingers have been involved (Thiemann 1909, Fleischner 1923, Kloiber 1926, Weil 1929, Dahs 1930, Dessecker 1930, Esau 1930), but patients have been described in whom nearly all the fingers and some of the toes were affected (Armstrong 1919, Shaw 1954).

The familial incidence of avascular necrosis of the phalanges has been noted by Armstrong (1919), Ryffel (1933), Sylvest (1940), Trippel (1950) and Shaw (1954). The mode of inheritance has been described as dominant, with strong penetrance. In one of our families twenty-three and in the other twenty persons were affected. It is clear that the appearance of the abnormality is controlled by a single autosomal gene which manifests itself in nearly all persons who are heterozygous for it. However, in one of our families there was a consanguineous marriage between affected persons. Two of the offspring of this marriage showed much more severe deformities than the rest, and it is suggested that they may have had a double dose of the abnormal gene. If this is so, the character cannot strictly be regarded as a " dominant," since the effects of the mutant gene in single and double dose are observably different. The general significance of this and similar observations in relation to the problem of dominance and recessivity of abnormal human traits is discussed elsewhere (Allison and Blumberg 1958).

CLINICAL MATERIAL

The material consists of two families showing what appears to be the same deformity. The proposita in the first family lives in a market town in Gloucestershire. Members of the family live in the neighbourhood, in Birmingham, and have emigrated to Australia, Canada and America. More complete information is available for this family and it will be presented in detail. Twelve affected members were seen either by the authors or by other physicians. Information on the remaining close relatives was obtained from several members of the family, cross-checked and confirmed by correspondence. The second family is from Berkshire, with some members resident in Wales. Five affected members of this family were seen, and the history of the others was obtained from relatives. There is no known relationship between the two families.

CASE REPORTS

FAMILY 1

Case 1—A woman aged forty-nine years (P in Fig. 4) was seen at the Radcliffe Infirmary, Oxford, in 1956 with the complaint of increasing thirst, hunger and urinary frequency, and was found to have diabetes mellitus, easily controlled by diet. There was no family history

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FIG. 1
Case 1—Photograph of the hands.

FIG. 2

FIG. 3

Case 1. Figure 2—Radiograph of right hand. Figure 3—Radiograph of right foot.

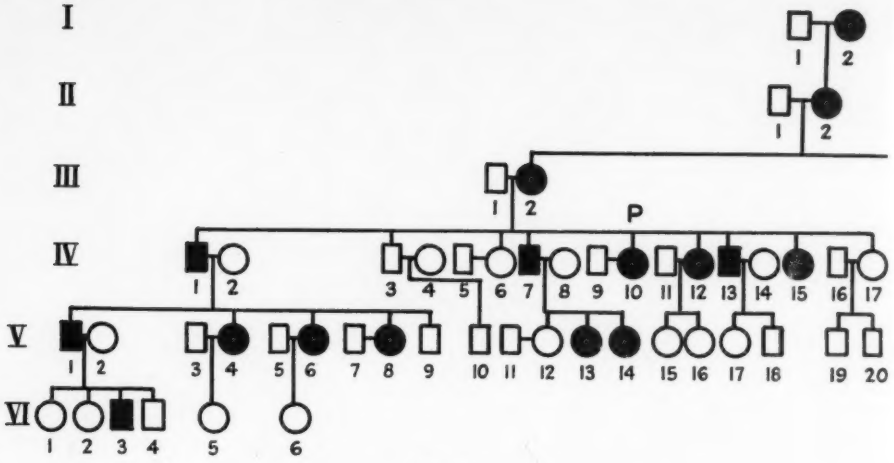

FIG. 4
Pedigree of family 1. ○=unaffected females; □=unaffected males;

of this disease. Changes in her hands were noticed as an incidental finding. She was said to have had " rheumatic fever " as a child, but remembered only that she had a febrile illness which kept her confined to bed for several weeks at about the age of ten years. She noticed occasional pains in her hands and wrists, usually when working in the cold. She had never been handicapped by the deformity; as a young woman she was able to play the piano, which she had since stopped for lack of interest. No other joints were involved. There was no history of relevant previous illness or occupational hazard.

On examination the general health appeared to be satisfactory, and there was no relevant abnormality in the respiratory, cardiovascular or neurological systems.

In the hands there was a nodular enlargement of the proximal and distal interphalangeal joints of all the digits (Fig. 1). Those of the terminal joints on all fingers (but not the thumbs) resembled Heberden's nodes. Movement at the distal interphalangeal joints of the fingers was limited to 10 degrees, and at the proximal interphalangeal joints to 90 degrees. Movement at the interphalangeal joints of the thumbs was 10 degrees. The terminal phalanges of the fingers were shortened. There was slight restriction of flexion at both wrists, but this was not accompanied by a deformity. There were no juxta-articular nodes. The feet showed hallux valgus. There was no deformity or restriction of movement in any other joint, nor any tenderness of joints or muscles. No tophi were present.

Investigations—The erythrocyte sedimentation rate was 11 millimetres in the first hour (Westergren), blood uric acid was 3·6 milligrams/100 millilitres. Haemoglobin and white cell content of the blood were normal.

Radiographic examination. Hands—The phalanges were small and stubby. The interphalangeal joints showed marked lipping and narrowing of the joint space with some flattening of the surfaces, as in osteoarthritis (Fig. 2). There were slight changes in the carpal bones. *Feet*—Both feet showed hallux valgus (Fig. 3). Changes like those in the fingers were seen in the joints of the great toe and, to a lesser extent, in the interphalangeal joints of the other toes. The other joints of the lower limb appeared normal, as did the spinal column and chest.

The family—The incidence of the digital abnormality in members of the family is shown in Figure 4, and a comparison of the involved and uninvolved members is shown in Table I. Several members of the family complained of intermittent pains in the hands and in the back, but there was no consistent difference from the clinical picture of the proposita. None had

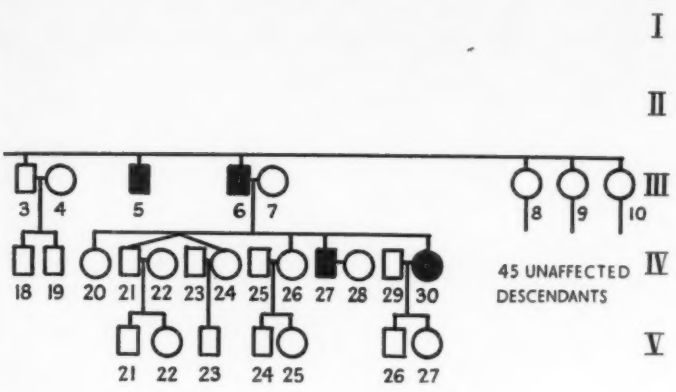

FIG. 4
●=affected females; ■=affected males; P=proposita (Case 1).

any other form of arthritis. All affected members of the family seen were functionally normal and many were employed in hard physical labour. The degree of involvement varied somewhat. A few subjects showed only slight abnormality, which might have been overlooked in any other family. All those affected stated that they had first noted the deformity at an early age (five to fifteen years) and that it had increased only slightly with the passage of time.

FAMILY 2

Case 2—The proposita in the second family was a woman aged sixty-one years (P in Fig. 7). In 1941 she was admitted to the Radcliffe Infirmary for treatment of a scalp wound. While she was being investigated, Dr A. M. Cooke noticed that her hands were deformed. She recalled that the joints of her fingers had been enlarged since she was at school, probably aged about twelve years. She had never had pain in the fingers or been inconvenienced by the joint enlargement, and could play the piano and do embroidery.

On examination the general health appeared good. In the hands there was a nodular enlargement of the proximal and interphalangeal joints of all the fingers (Fig. 5). The terminal phalanges were shortened. Movement was limited to about 50 degrees in the proximal joints and 15 degrees in the distal joints. The enlargement of the joints was firm and bony, and covered by normal skin. There were no tophi, and the other joints were clinically normal.

Investigations—The erythrocyte sedimentation rate was 8 millimetres in the first hour (Westergren). The blood uric acid was 2·8 milligrams/100 millilitres. Haemoglobin and white cell content of the blood were normal.

Radiographic examination. Hands—The changes were like those in Case 1, with involvement of the metacarpo-phalangeal joints of all the digits (Fig. 6). All the phalanges were shortened. Both wrists were involved.

Case 3—A male cousin of the patient in Case 2 (IV, 14 in Fig. 7) was seen in 1954, when he was aged forty-seven years. He had marked deformities of both hands, which had been present since he was a young child. The phalanges were all much shortened, with marked bony thickening at the interphalangeal joints of all fingers and of both thumbs; there was also slight thickening of the metacarpo-phalangeal joints. The range of movement at the metacarpo-phalangeal joints was nearly normal, but was restricted to about 20 degrees at the proximal interphalangeal joints and less than 10 degrees at the distal interphalangeal

FIG. 5
Case 2—Photograph of the hands.

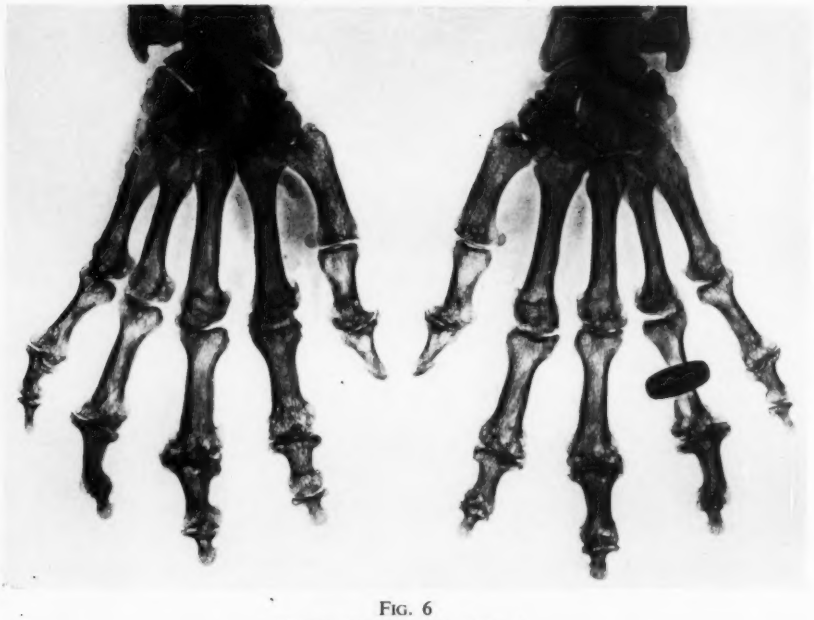

FIG. 6
Case 2—Radiographs of hands.

joints. The patient had had no pain in the hands but was conscious of some functional disability. He had been in good health. He failed to keep an appointment for full examination, and returned to Wales. Two years later he was reported to have died, the cause of death being unknown.

As shown in Figure 7 this patient was the offspring of a first cousin marriage between two affected persons. He was much more severely affected than his brother (IV, 13 in Fig. 7)

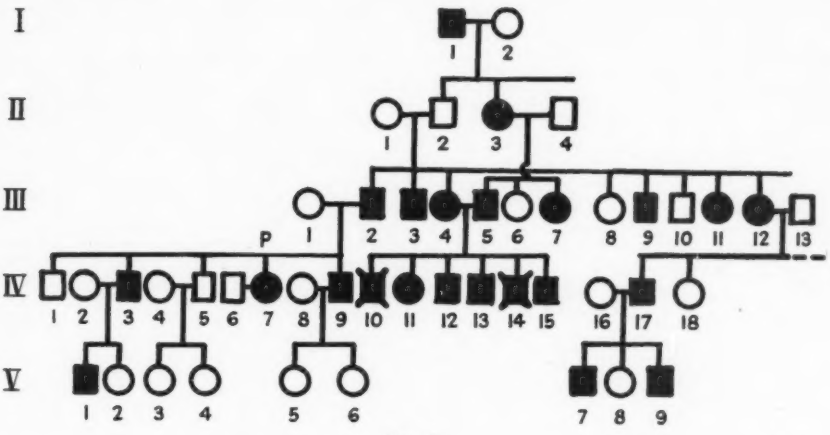

FIG. 7

Pedigree of family 2. ○ = unaffected females; □ = unaffected males; ● = affected females; ■ = affected males; ⧓ = marked involvement; P = proposita. Note the consanguineous marriage of involved cousins and their offspring in generation III, IV, and the greater degree of involvement of two of the offspring.

and other members of the family examined by one of the authors. An elder brother (IV, 10 in Fig. 7) was reported to have been severely affected, but he had died at the age of fifty-six from coronary thrombosis before the present investigation began.

DIFFERENTIAL DIAGNOSIS

The condition we have observed can be distinguished from osteoarthritis, rheumatoid arthritis and gout by its early age of onset, equal sex incidence, benign course, absence of symptoms, characteristic joint distribution, lack of abnormal laboratory findings, and its well defined genetic control. There is some resemblance to primary generalised osteoarthritis (Kellgren and Moore 1952), but signs of the latter begin later in life, and usually show less symmetrical affection, without shortening of the phalanges. The deformity of the distal interphalangeal joints in our cases resembled that of Heberden's nodes, but the accompanying changes in the proximal interphalangeal joints, the early age of onset and equal sex incidence mark the condition as distinct. The benign course and lack of general skeletal involvement differentiate it from osteoarthritis deformans endemica (Goldstein and Nikiforov 1931) and from Morquio's disease (Ritvo 1955).

DISCUSSION

The condition we have observed appears to be one of a group of diseases characterised by avascular necrosis of the epiphyses. The pathological changes shown by similar patients were described by Dessecker (1930) who concluded from histological examination that it was an avascular necrosis, thus confirming the suggestions of radiologists. The epiphyses appear to become damaged by compression and splitting, and defects occur as a result of

absorption of bone. The growth of the phalanx is retarded and the epiphysis and metaphysis are widened. Later, there is a regeneration of the epiphysis which joins with the metaphysis at about the time of puberty. The etiology and pathogenesis are unknown, but presumably the initiating stimulus and the localisation of the defect to a particular joint are genetically determined. Occasionally, more than one site may be affected: Ryffel (1933) described a case in which Perthes' disease of the right femoral head was observed in a five-year-old boy; five years later the patient had avascular necrosis of the phalanges of both hands. The average age of onset in our cases was before puberty, which agrees with most of the other reports.

TABLE I

THE OFFSPRING OF AFFECTED INDIVIDUALS (NUMBER AT RISK) AND THE NUMBERS AFFECTED IN EACH FAMILY
(The offspring of the cousin marriage in family 2 are excluded. Only the sub-families for which complete information is available are included in family 2)

Family	Number at risk			Number affected			Percentage involved		
	Males	Females	Total	Males	Females	Total	Males	Females	Total
Family 1	17	25	42	9	9	18	52·9	36·0	42·9
Family 2	11	7	18	7	4	11	63·6	57·1	61·1
Both	28	32	60	16	13	29	57·1	40·6	48·3

The mode of inheritance of the condition in both families resembles that previously described by Armstrong (1919) and others (Ryffel 1933, Sylvest 1940, Trippel 1950, Shaw 1954). From the compiled results in Table I it is clear that the condition has appeared in our families in nearly half the offspring of matings in which one partner had it. Thus it seems that the appearance of the abnormality is controlled by a single gene segregating in these families, which has nearly complete penetrance in the heterozygous phase. If the report is correct one carrier of the gene in the second family showed no defect. The expression of the gene varies somewhat, but in general gives rise to only a relatively slight deformity. The presence of two severely affected persons among the offspring of the consanguineous mating in family 2, however, suggests that they may represent the effects of the abnormal gene in double dose. If this is so, the type of inheritance cannot strictly be regarded as " dominant," because the abnormal homozygote is different from the heterozygote. In a number of other instances, when it has been possible to compare persons homozygous for abnormal genes with those showing heterozygous effects, the two have been found to be different (Allison and Blumberg 1958). It therefore seems that human abnormalities very rarely show the typically dominant type of inheritance.

SUMMARY

1. Two unrelated families with a genetically determined arthritis-like syndrome of the joints of the hand and wrist are described.

2. The condition begins before puberty, is relatively painless and is not functionally disabling. The condition resembles that described by Thiemann and appears to fall into the group of avascular necrosis.

3. The appearance of the abnormality is controlled by a single autosomal gene which manifests itself in nearly all persons who are heterozygous for it. Two of the offspring of a marriage between affected persons showed much greater involvement than their siblings.

This suggests that they may represent the effect of the abnormal gene in double dose. If this is so, the type of inheritance cannot strictly be regarded as " dominant," since the abnormal homozygote is different from the heterozygote.

The authors are much indebted to Dr A. M. Cooke and Dr D. Pyke for bringing to their attention cases admitted to the Radcliffe Infirmary, Oxford, under the care of the former. Dr Henry Burger, of the Royal Australian Air Force, kindly examined two of the patients and provided us with photographs and radiographs.

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HYPERPARATHYROIDISM DUE TO PARATHYROID CARCINOMA

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Carcinoma of a parathyroid gland is an uncommon cause of hyperparathyroidism. Black (1954) was able to collect twelve cases of parathyroid carcinoma with hyperparathyroidism and distant metastasis. He considered that another group of four cases undoubtedly represented carcinoma, because a tumour found to be densely adherent or locally invasive at operation recurred later. Cope, Nardi and Castleman (1953) reported four cases in a series of 148 cases of hyperparathyroidism. Three of the patients were found to have metastases at necropsy, but in the fourth neither recurrence nor hyperparathyroidism had occurred four years after removal. Wray (1953) reported a further case in which the tumour was found to be locally invasive at operation. The growth recurred and proved to be inoperable because of infiltration. Although distant metastases were not present at necropsy, the malignant nature of the tumour is beyond doubt. Recently Weissman, Worden and Christie (1957) reported a case of mediastinal parathyroid carcinoma which, three years after surgical removal, recurred with multiple metastases and hyperparathyroidism. Thus the sixteen cases reported by Black, the three cases of Cope, Nardi and Castleman, Wray's case, that of Weissman, Worden and Christie, and the case here reported, bring the total of indisputable functioning parathyroid carcinomata to twenty-two.

CASE REPORT

The patient, a married woman of fifty-six, noticed a lump in the right upper jaw in July 1955. This was curetted, and proved to have the histology of an osteoclastoma. Further investigation showed that the serum calcium was increased and serum inorganic phosphate diminished. On these findings a diagnosis of hyperparathyroidism was made. Seven years before, in 1948, she had had five stones removed from the right kidney, followed by right nephrectomy for non-functioning kidney associated with hypertension. She had suffered from recurrent bouts of dyspepsia since adolescence. Treatment with diet and alkalies had produced symptomatic relief of variable duration.

Physical examination revealed, behind the sterno-clavicular joint, a small mobile swelling which moved on swallowing. Several soft, smooth tumours of the arms and axillae were thought to be neurofibromata. The blood pressure was 240/130 millimetres of mercury; there was no clinical evidence of left ventricular hypertrophy or heart failure.

Investigations (November 1955)—The serum calcium was 12·8 milligrams/100 millilitres; the serum inorganic phosphate 2·2 milligrams/100 millilitres; alkaline phosphatase 32·5 K.A. units; urinary calcium excretion 312 milligrams in twenty-four hours; blood urea 54 milligrams per cent; serum proteins and electrolytes were normal.

Operation—Mr R. Marnham explored the neck and removed a tumour about two centimetres in diameter in relation to the lower pole of the left lobe of the thyroid. The tumour was adherent and difficult to remove. No local metastases were discovered.

Histology—Histological examination revealed parathyroid tissue with mainly clear cells. There was evidence of capsular invasion and there were large numbers of nuclear mitoses. On these grounds a diagnosis of parathyroid carcinoma was made. After operation there was a fall in serum calcium with tetany (Fig. 1).

Further treatment and progress—Calciferol was given for the next five months (50–100,000 units daily). It was hoped that this would assist in the maintenance of the serum calcium

level and minimise any stimulus to hypertrophy falling upon remaining parathyroid tissue. The patient was without symptoms until August 1956, when she complained of increased urinary frequency, extreme tiredness and vague pains in the back and legs. She was admitted to hospital for further study.

Investigations (November 1956)—The serum calcium was 13·7 milligrams/100 millilitres; serum inorganic phosphate 2·4 milligrams/100 millilitres; serum alkaline phosphatase 10·7 K.A. units; plasma proteins total 6·2 grammes (albumin 3·7 grammes; globulin 2·5 grammes; albumen-globulin ratio 1·5); plasma chloride 106 m.eq./l; plasma sodium 139 m.eq./l; plasma CO_2 combining power 24·5 m.eq./l; blood urea 47 milligrams; urea clearance test 38 per cent of average normal; blood count Hb. 93 per cent (13·8 grammes), P.C.V. 41 per cent; erythrocyte sedimentation rate 7 millimetres/1 hour (Wintrobe); fractional test meal—marked hyperchlorhydria (free HCl≡84 millilitres N/10 HCl in one specimen).

FIG. 1

Diagram showing the variations in serum calcium (solid line) and serum inorganic phosphate (interrupted line). The values are expressed as milligrams/100 millilitres. The fall in serum calcium after the first operation is well shown. A similar result does not occur after the second operation.

Radiographic examination—Skeletal survey showed generalised osteoporosis. The trabecular pattern in several bones was coarsened. There was slight cortical absorption in the phalanges as evidence of active hyperparathyroidism. The skull was normal.

Intravenous pyelography showed poor excretion from the remaining (left) kidney. Barium meal showed some persistent tenderness over the duodenum, which was deformed. The appearances suggested duodenal ulcer.

Electrocardiographic studies were within normal limits except for some shortening of the QT interval.

Balance studies—Balance studies were carried out for six periods of six days. The diet was constant with a calcium intake of 910 milligrams a day. Faecal and urinary estimations of calcium, phosphorus and nitrogen were made. The patient was in a strongly negative balance for calcium. Average urinary calcium was 400 milligrams a day and faecal calcium 900 milligrams a day.

Further course—An attempt was made to treat the patient with oral phosphate. (The dosage of di-sodium hydrogen phosphate was equivalent to 2 grammes of phosphorus a day.) It was hoped that this might raise the serum phosphorus and produce a secondary reduction in serum

calcium. In fact, serum inorganic phosphate increased from 3 milligrams to 5·5 milligrams/100 millilitres while the serum calcium continued to increase from 15 to 17 milligrams per cent. Phosphates caused diarrhoea and were discontinued. Balance studies while phosphate was being given showed a marked increase in faecal calcium (2·4 grammes a day) with little change in urinary calcium. A second exploration of the neck was carried out by Mr Marnham in December 1956 with the hope that any functioning tumour tissue might be removed in order to prolong life. Multiple grey, firm nodules about three millimetres in diameter were removed from the left side of the neck. A remnant of thymus containing two firm nodules was also removed. Histological examination of the nodules showed areas of clear cell tumour tissue, confirming the diagnosis of parathyroid carcinoma.

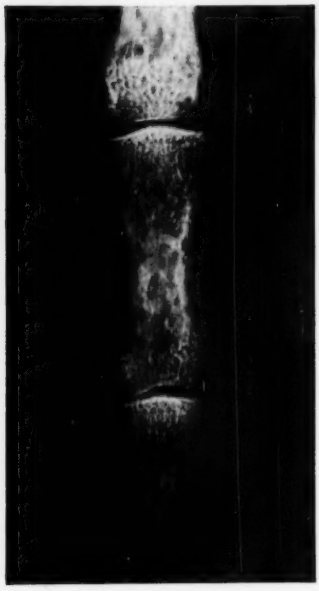

FIG. 2
Radiograph of the left ring finger showing decalcification and cortical absorption; evidence of active hyperparathyroidism.

The patient recovered from the operation but there was no improvement. There was no post-operative tetany, nor any lowering of the serum calcium (Fig. 1). During the next few weeks the blood urea rose to 108 milligrams/100 millilitres, but fell to 56 milligrams/100 millilitres under the influence of a low protein, high calorie diet.

Professor Charles Dent suggested that an attempt to lower the serum calcium might be made with methyl thiouracil. Some evidence has been obtained (C. E. Dent, K. E. Halnan and E. E. Pochin, unpublished observations) that added thyroxin potentiates the effect of parathormone on the serum calcium level and that hypothyroidism decreases its action. In the period that the drug could be given (four weeks; 400 milligrams daily) no benefit was apparent, and it did not appear to influence serum calcium levels. As the patient's condition was deteriorating, cortisone was given (60 milligrams daily—total 960 milligrams) in the hope that it might have an anti-parathyroid effect, but its administration was also without benefit.

By this time (six weeks after the second operation) definite radiological evidence of hyperparathyroidism had developed. Cortical absorption was visible in the phalanges, with porosis and pseudo-cysts (Fig. 2). The patient died thirteen months after the diagnosis had been confirmed at the first operation.

Necropsy findings—Necropsy revealed the following significant findings. 1) Several minute grey nodules in the neck shown by histological examination to be carcinomata of the parathyroid gland (Figs. 4 and 5); 2) the right lobe of the thyroid was intact and two parathyroid glands of normal size were demonstrated on its postero-lateral surface; 3) a metastatic deposit in the lung (Fig. 6); 4) calcinosis of the remaining (left) kidney; 5) some metastatic calcification of the cranial dura mater; 6) cardiac hypertrophy (weight 460 grammes) with scattered plaques in the ventricular myocardium due to calcinosis; 7) a healed peptic ulcer on the posterior wall of the pylorus.

DISCUSSION

In benign hyperparathyroidism the histological appearances sometimes misleadingly suggest early carcinoma. The diagnosis of parathyroid carcinoma is established beyond all doubt if the tumour is associated with hyperparathyroidism and distant metastasis. If the tumour is locally invasive and recurs after surgical removal it may also be accepted as

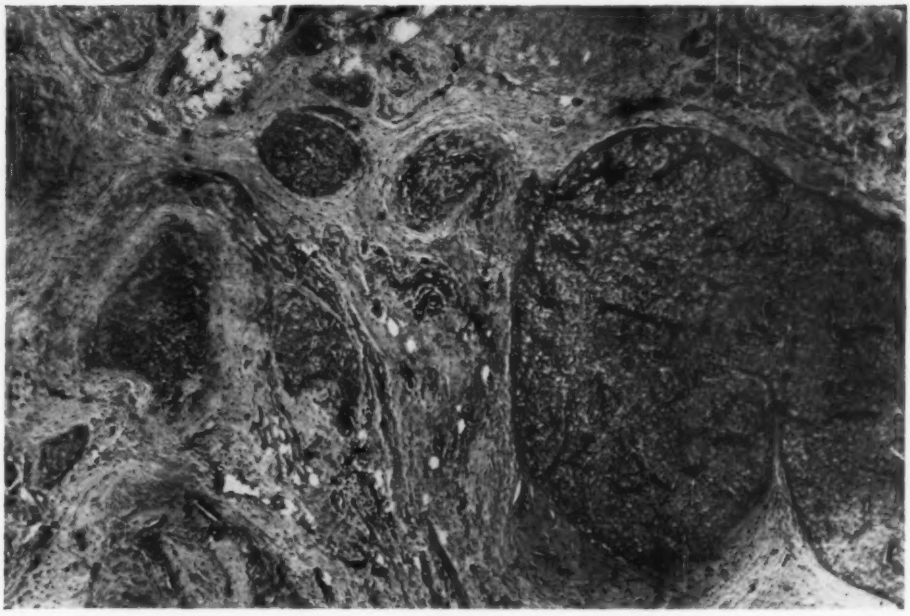

FIG. 3
Low power view of recurrent parathyroid tumour. Note nodular masses of tumour cells with infiltration of surrounding tissue. (×40.)

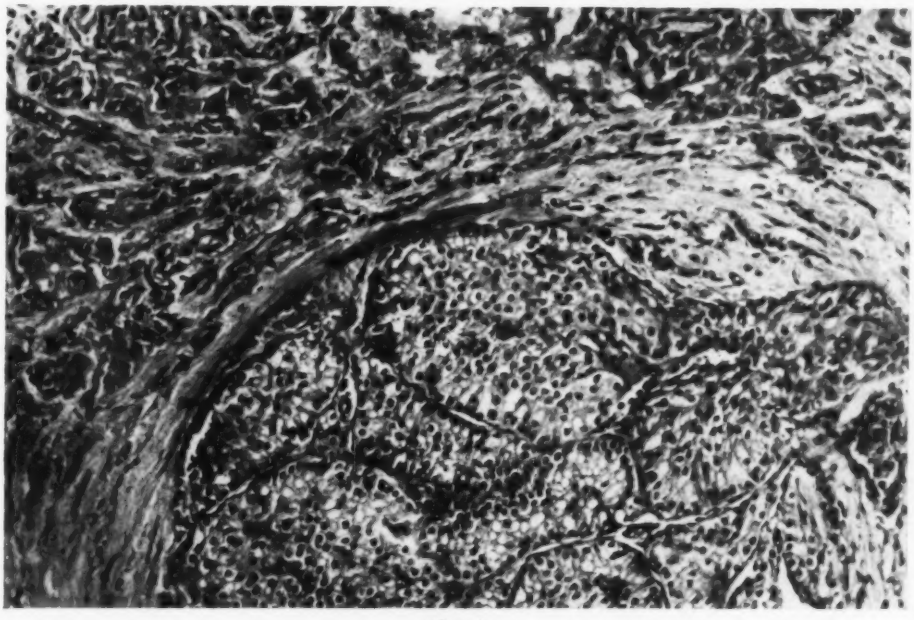

FIG. 4
Higher magnification of recurrent tumour.

carcinomatous. This paper concerns a case with hyperparathyroidism, local recurrence after surgery and metastasis. Although non-functioning parathyroid carcinoma probably exists, it is a less definite entity.

The most common cause of death in carcinoma of the parathyroid with hyperparathyroidism is renal failure. There is no significant difference in the sex distribution, and the disease is commonest in the fifth decade.

Castleman (1952) based the pathological diagnosis of carcinoma upon evidence of capsular invasion, local infiltration, loss of the normal parathyroid pattern and a trabecular arrangement of the cells. The cells themselves are often cylindrical, and numerous mitoses are present. The nuclei are deeply staining and enlarged nucleoli are often visible. Tumour cells within blood vessels also support the diagnosis of malignancy, but are not confined to malignant tumours. In this case the tissues removed at both operations and from the neck and lung at

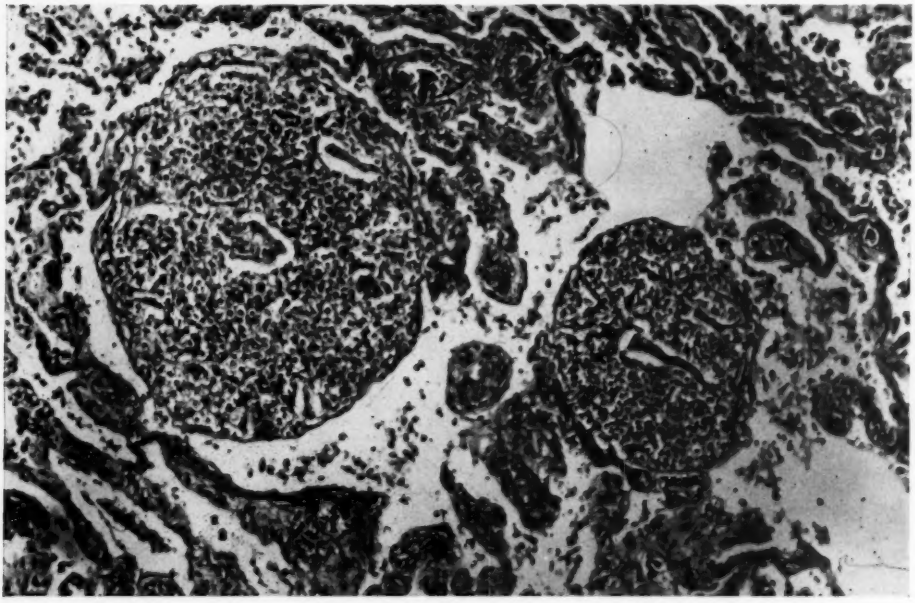

FIG. 5
Two microscopic nodules of tumour tissue in the lung.

necropsy fulfil these criteria. The tumours are locally invasive, which renders surgical removal difficult. Often this may be the first signal that the tumour is not a simple parathyroid adenoma. Regional lymph nodes and the liver are common sites for metastasis.

Treatment of the condition is unsatisfactory. If a carcinoma is suspected at operation a wide excision is indicated, together with a thorough search of the neck for lymphatic involvement or secondary nodules. The improvement after surgery commonly disappears with the onset of local recurrence or metastasis. A second operation under such circumstances is technically difficult, and in this case, as in others, was unsuccessful. Black (1954) makes a plea for post-operative irradiation, but any improvement from this form of treatment seems to be partial and temporary.

Treatment was attempted with oral phosphates, cortisone and methyl thiouracil without success. Radical surgical removal of neoplastic parathyroid tissue remains the only form of treatment with any hope of cure or improvement.

A further point of interest is the association between peptic ulcer and hyperparathyroidism reported by several authors (Rogers 1946, *Lancet* annotation 1955). In this case there was marked hyperchlorhydria, and the scar of a healed duodenal ulcer was demonstrated at necropsy. The symptoms of peptic ulcer had been present for more than thirty years. It is difficult to define the onset of hyperparathyroidism, but renal stones were removed nine years before death.

SUMMARY

A case of hyperparathyroidism from carcinoma of the parathyroid gland which recurred locally and metastasised to the lung is described. There was a history of renal lithiasis and peptic ulceration. Improvement followed initial surgical removal but the tumour recurred and death resulted from hyperparathyroidism.

My thanks are due to the staff, both nursing and technical, of the metabolic unit of the Institute of Orthopaedics and the Royal National Orthopaedic Hospital where this patient was treated. I am indebted to Dr H. A. Sissons and Dr P. G. Walker for the pathological investigations and to Professor Charles Dent for helpful advice.

This patient was under the care of Dr Reginald Nassim, and I am especially grateful to him both for guidance and for permission to publish this paper.

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FURTHER OBSERVATIONS ON THE ELASTIC MECHANISM OF THE INTERVERTEBRAL DISC*

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In the light of fresh experimental evidence (Happey, Horton, MacRae and Naylor 1955) the elastic mechanism of the intervertebral disc can now be enlarged upon and attention drawn to the relation between decrease in height of the disc and increase in circumference of the annular wall.

The present investigation was made with a polarising microscope and a comparatively high intensity x-ray beam, enabling the orientation of the annulus at fibre and protofibrillar levels to be investigated. Figures 1 and 2 are typical x-ray photographs obtained from thin vertical sections of undried and air-dried material. The corresponding photomicrograph is shown in Figure 3. The disorientation of the protofibrils caused by the drying of specimens can clearly be seen in Figures 1 and 2.

It was thought that the biaxial x-ray diagram might arise from inclination of alternate layers of collagen fibres (termed " uniaxial sheets ") at an angle Ω to each other, the fibres in each sheet being approximately parallel to each other. This was confirmed as follows.

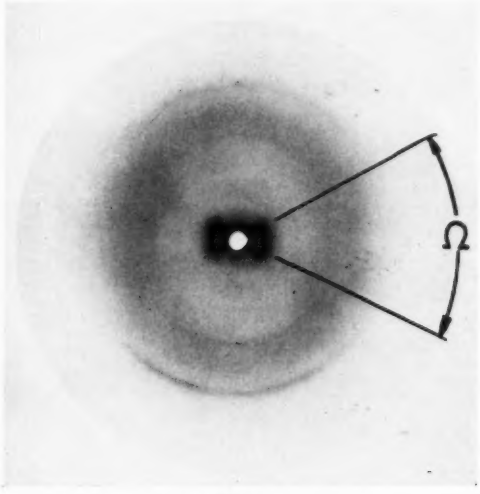

FIG. 1

Annulus fibrosus, age twenty-five years. Undried specimen showing biaxial arrangement of collagen protofibrils.

Fresh cadaveric discs from necropsy subjects were divided horizontally, exposing both the annulus fibrosus and nucleus pulposus. The material was frozen to about −80 degrees centigrade with solid carbon dioxide, then cut vertically with a sharp razor blade, yielding sections which, when dried in the atmosphere, were about 10μ thick. In this manner individual collagen sheets were dissected from the annulus of intervertebral discs of subjects aged from

* This work forms part of a thesis submitted to the University of London for the degree of Doctor of Philosophy.

twenty-five to seventy-seven years. These sheets were shown to have a uniaxial orientation by polarising microscope and x-ray observations (Figs. 4 and 5).

The following experimental arrangement was used in order to demonstrate that in the annulus fibrosus the fibres, as observed under the polarising microscope, are parallel to the direction of the protofibrils as revealed by x-ray analysis. In general two sets of fibres, which

FIG. 2
Same specimen, air-dried, showing equatorial arcs coalescing into single arc.

correspond to the two collagen orientations, may be detected by means of the polarising microscope in the thicker specimens used in x-ray analysis.

A thin section of annulus fibrosus about 5μ thick, sectioned as described earlier, was mounted on a brass specimen holder and a hair was mounted under the polarising microscope, across the section, so as to be approximately parallel to the fibres. A photomicrograph was

TABLE I

Ω (x-rays)	Angle between fibres of consecutive sheets
53	—
60	59
59	62
70	68
75	73
52	48
60	57
59	57
59	57
59	57

taken of the fibres and the hair. While a radiograph was being taken of the area under examination, the outer side of the wire rectangle rested against the x-ray casette and so left a shadow parallel to the fibre direction. By this means it was shown that the protofibrils, whose directions are perpendicular to the 11·6A degrees reflections in the x-ray diagram, correspond to the fibres directions observed under the polarising microscope.

It therefore seems quite clear that the biaxial x-ray diagram arises from the superposition of two sets of collagen fibres mutually inclined at an angle Ω to each other. Added confirmation was obtained by observing the angle between the protofibrils of adjacent sheets as measured

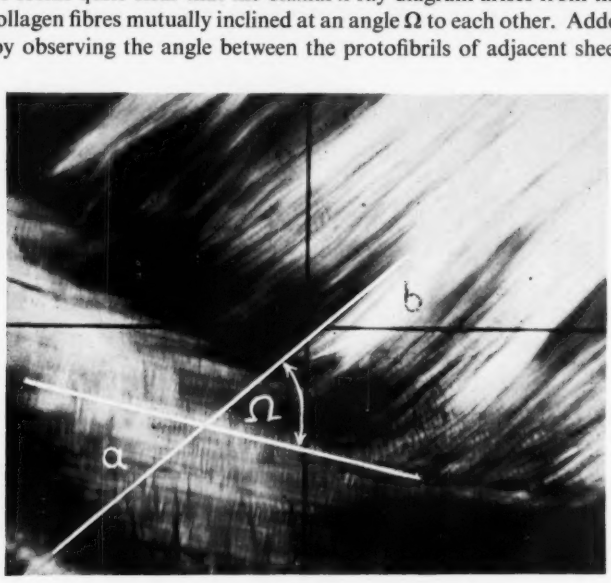

FIG. 3
Photomicrograph using polarised light showing the biaxial orientations of the fibres. The mutual orientation of the fibres is at an angle Ω in the plane of the disc wall. This orientation is also shown in Figure 1 by the angle Ω between the equatorial reflections in the x-ray diagram.

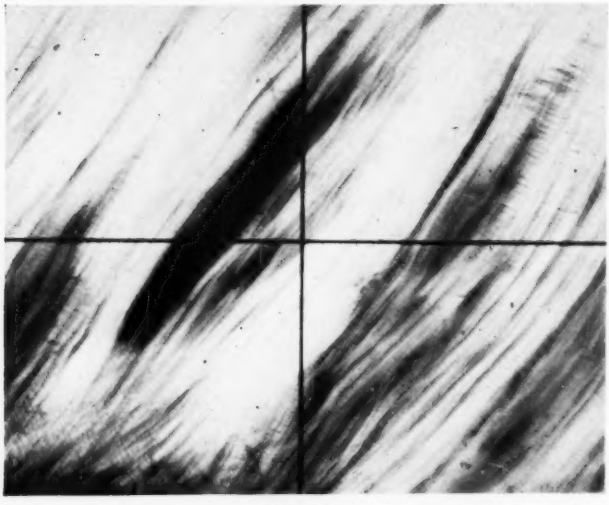

FIG. 4
Photomicrograph of thin section of annulus showing the uniaxial orientation of the fibres.

from x-ray diagrams and comparing this with the angle between the fibres of the uniaxial sheets as seen under the polarising microscope. As shown in Table I, good agreement was obtained between the two methods.

It was of interest to obtain some idea of the regularity in thickness and orientation of the uniaxial sheets in relation to each other. For this purpose, discs were sectioned horizontally and the sections examined under the polarising microscope. The thickness of individual sheets was examined at regular intervals round the annular wall, as was the thickness of consecutive sheets. Table II illustrates the type of result obtained.

It is apparent from the above table that quite large variations in thickness occur within a given uniaxial sheet, and, furthermore, there are large and apparently irregular differences in the thicknesses of these sheets.

It would appear that growth in the annulus fibrosus consists not of an increase in the number of uniaxial sheets present in the annulus, but of a thickening of the uniaxial sheets developed in early life. However, three discs obtained from older subjects have shown in the nucleus pulposus a typical cross structure that we have come to associate with annulus fibrosus, implying proliferation of the annulus fibrosus into the nucleus pulposus.

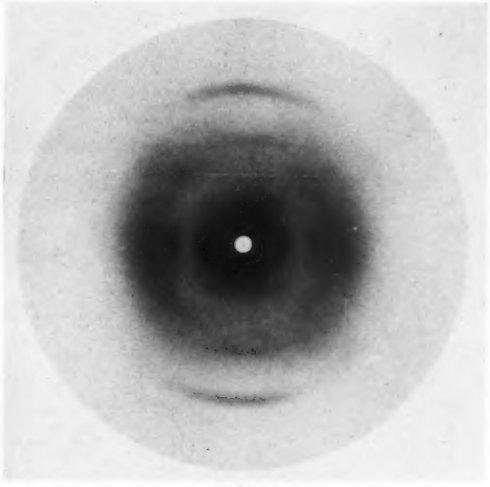

FIG. 5

X-ray photograph of an undried section of annulus shown by polarising microscope observations to have uniaxial orientation of fibres.

The arrangement of these uniaxial sheets in relation to each other is very regular. In all specimens examined, the pattern was of the type *a*, *b*; *a*, *b*; *a*, *b*; etc., where *a* and *b* are taken to represent the orientation of two consecutive uniaxial sheets mutually inclined at an angle Ω to each other. The only irregularity, as already noted, was in the thickness of the sheets.

DISCUSSION

Previous work (Happey, Horton, MacRae and Naylor 1955) has drawn attention to the ability of the nucleus pulposus of an intervertebral disc to convert a vertical pressure into a horizontal thrust on the annulus fibrosus. Furthermore, it has been suggested that the annulus fibrosus must be elastic in order to dissipate the resultant thrust from the nucleus. Any proposed elastic mechanism must provide for an increase in the radius of the disc, the distance between the cartilage end plates decreasing with increase of cross-sectional area. It is suggested that such an increase in radius can be achieved by the movement of adjacent uniaxial sheets, with consequent decrease in the value of Ω as outlined in Figure 6. The proposed mechanism also accounts for the flexibility of the spine.

Only limited angular changes in Ω are possible because of the strong cohesion between adjacent sheets; under laboratory conditions reversible changes of up to 15 degrees have been recorded. The approximate increase in circumference is given by

$$E=\frac{\cos \frac{1}{2}\Omega_2}{\cos \frac{1}{2}\Omega_1}$$ where Ω is the angle between successive sheets and $\Omega_1>\Omega_2$.

It follows from this equation that for large values of Ω a given change of angle will result in a greater increase in circumference than for those parts of the annulus where Ω is comparatively small. Hence a uniform pressure from within the centre of the disc would probably cause a localised bulging in such places rather than at other sites.

TABLE II

Specimen 1		Specimen 2	
Sheet number	Thickness (*millimetres*)	Sheet number	Thickness (*millimetres*)
Subject aged 57 years (specimen undried)			
1	1·05	1	0·40
2	0·75	2	0·70
3	0·95	3	0·40
4	1·35	4	0·50
5	0·80	5	0·30
Subject aged 67 years (specimen air-dried)			
1	0·15	1	0·10
2	0·20	2	0·15
3	0·05	3	0·30
4	0·15	4	0·10
5	0·10	5	0·20
6	0·15	6	0·15
7	0·10	7	0·30
8	0·15	8	0·50
Subject aged 21 years (specimen undried)			
1	1·00	1	1·25
2	0·90	2	1·10
3	0·80	2	0·95
4	0·90	4	1·35
5	2·70	5	1·56
6	2·65	6	0·60

Assuming, as a first approximation, that an adult lumbar disc is a cylinder of radius 2·5 centimetres, it is possible to calculate the increase in circumference and hence change in Ω from the observations reported by Hirsch and Nachemson (1954). They found that the increase in radius when an additional 60 kilograms were added to a disc already compressed by 40 kilograms was 0·2 millimetres.

Circumference of the disc before loading with 60 kilograms $=2\pi r=2\pi\times 2{\cdot}5=5\pi$ centimetres.
Circumference of the disc after loading with 60 kilograms $=2\pi(r+dr)=2\pi\times 2{\cdot}52=5{\cdot}04\pi$ centimetres.
$\therefore$ Change in circumference $=2\pi\times 0{\cdot}02=0{\cdot}04\pi$ centimetres.

$$\text{Hence percentage change in circumference}=\frac{0{\cdot}04\pi}{5\pi}\times 100=0{\cdot}8 \text{ per cent.}$$

For a value of $\Omega=50$ degrees, a change of 1 degree would give the necessary change in radius, indicating that only small changes in the annular wall are necessary to accommodate the stresses to which it is normally subjected.

Finally, the results from the investigation of undried specimens indicate that once the annulus fibrosus has reached maturity its structure does not change, though there may be

some reduction in cohesion between the uniaxial sheets in old age, leading to a weakening of the annular wall. With increasing age the efficiency of the intervertebral disc is impaired and this can probably be attributed to the changes occurring in the nucleus pulposus, principally the loss of its gel properties together with the deposition of large oriented bundles of fibres. At this stage it has lost the ability to transmit pressure evenly to all parts of the annulus

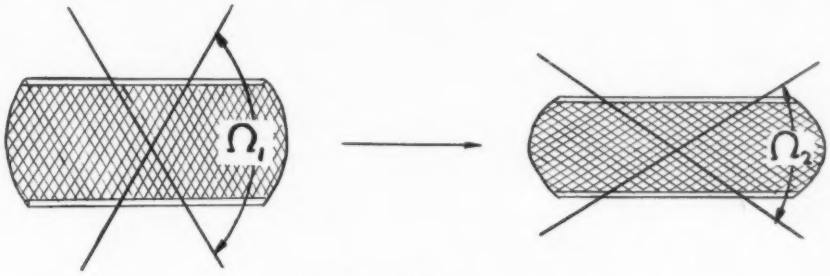

FIG. 6

Proposed elastic mechanism for the annulus fibrosus. Vertebral pressure causes a decrease of Ω from Ω_1 to Ω_2, leading to an increase in the circumference of the disc.

fibrosus. It has been suggested (Happey, MacRae and Naylor 1953) that the loss of resiliency caused by such biochemical changes may be one of the factors underlying the loss of physiological function of the disc with advancing years, with secondary effects upon the associated spinal joints. In particular, the mobility of the joints may be restricted.

SUMMARY

By polarising microscope and x-ray crystallographic techniques the annulus fibrosus has been shown to consist of regularly oriented sheets of collagen fibres. These results have been interpreted in terms of an elastic mechanism whereby thrust from the nucleus causes increased girth in the annulus. It is suggested that this is accomplished by a change in the angle between the axis of the fibres in adjacent uniaxial layers of the annulus. Furthermore, the loss of elasticity of the intervertebral disc associated with age would seem to be mainly due to changes occurring in the nucleus pulposus.

It is a pleasure to thank Dr F. Happey, Head of the Textile Department, Bradford Technical College, for his help and guidance; and Mr A. Naylor, Consultant Orthopaedic Surgeon to the Bradford Hospitals, for helpful discussion and specimens.

Figures 1 to 4 are reproduced by courtesy of the Editors of *Nature*.

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THE INFLUENCE OF CORTISONE AND A.C.T.H. ON BONE SUBJECTED TO MECHANICAL STRESS (TOOTH MOVEMENT)

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Mechanical stress is known to have significant effects on connective tissues generally, and consequent bone changes are of everyday importance. Knowledge of these is dependent on our obtaining detailed and accurate data. The precise measurement of stresses often presents experimental difficulties, but manipulation of the teeth allows exact and delicate observations, whether brief or prolonged.

Bone changes associated with mechanically produced tooth movement have been studied in man, the rabbit, the guinea pig and the rat, where the influence of variables such as the degree and duration of the applied force, age, sex and menstrual cycle were determined (Smith and Storey 1952; Storey 1954, 1955*a*, *b*, *c*). The experimental method, adapted to animals, gives a unique opportunity for exploring details of bone alterations under controlled pressures and tensions. It has now been extended to include the effect of various hormones on bone when subjected to these stresses. This present paper is concerned with the effect of cortisone and A.C.T.H. in the rabbit and guinea pig.

EXPERIMENTAL METHOD

Helical torsion springs were applied to the incisor teeth of rabbits and guinea pigs in order to move them laterally. These springs consist of a number of loops of wire wound into two central coils, and two free arms. The arrangement and shape of the springs is shown in Figures 1 and 2, and their dimensions are given in Table I; the arms of the springs are inserted into fine holes drilled through the crowns of the teeth. The evolution of suitable springs has been discussed before (Smith and Storey 1952).

TABLE I

DIMENSIONS OF AUSTENITIC STAINLESS STEEL SPRINGS USED IN THE EXPERIMENTS ON THE RABBIT AND GUINEA PIG

	Rabbit	Guinea pig
Length of arm . (*inches*)	0·425	0·20
Number of coils . . .	19	19
Diameter of wire (*inches*)	0·0120	0·010

TABLE II

THE NUMBER OF ANIMALS USED IN EACH EXPERIMENT

		Normal	Treated with cortisone	Treated with A.C.T.H.
No mechanical stress	Rabbit .	3	4	4
	Guinea pig	3	4	4
Mechanical stress .	Rabbit .	8	8	6
	Guinea pig	7	14	7

The initial force applied to the incisor teeth of both the rabbit and the guinea pig was of the order of 50 grammes. An initial deflection of each arm of the spring through 90 degrees of arc was considered essential in order to obtain a steep deflection-load curve and so maintain

FIG. 1

FIG. 2

The experimental arrangement of the helical torsion springs on the upper incisor teeth of the rabbit. Figure 1—Before tooth movement. Figure 2—During tooth movement. (×3.)

the force at a reasonably constant level over the small range of tooth movements: as the incisor teeth moved apart there was only a small decrease in the applied force—in the rabbit, 7·5 millimetres tooth movement, a drop from 50 to 40 grammes, in the guinea pig from 50 to 25 grammes. The method of measurement has been described before (Storey 1955*a*).

Guinea pigs (eight to twelve weeks old) were killed at daily intervals for seven days and rabbits (eight to twelve weeks old) at two, four, seven and eleven days.

Table II shows the six groups of animals used in these experiments.

Hormones—Cortisone acetate (Merck) was administered subcutaneously, 25 milligrams a day to rabbits and 5 or 10 milligrams a day to guinea pigs. Long acting A.C.T.H. was administered similarly—10 international units a day to rabbits and 3 international units a day to guinea pigs.

Histological sections—The upper incisor teeth and surrounding bone were dissected from the animal and decalcified in 5 per cent nitric acid and then embedded in paraffin wax. Sections

FIG. 3

FIG. 4

Figure 3—Photomicrograph of the premaxilla of the normal rabbit showing the relationship of the upper incisor teeth to the premaxillary bones and suture. (×10.) Figure 4—Photomicrograph of the premaxilla after seven days' tooth movement. The premaxillary bones as well as the teeth have moved laterally, and new bone laid down on the ends of the bony serrations has maintained the continuity of the suture. New bone trabeculae have formed, orientated in the direction of movement of the teeth, at the peridental membrane bone junction. (×10.)

were cut through the incisor teeth at an angle of 20 to 25 degrees to the coronal plane and included incisor teeth and premaxillary bone on both the medial and lateral sides of the teeth. These sections were cut at 10μ and stained with Ehrlich's acid haematoxylin and eosin.

RESULTS

THE RABBIT

No mechanical stress. *Normal rabbit*—In the rabbit aged eight to twelve weeks the two premaxillary bones are joined at the intermaxillary suture, where serrations of bone interdigitate and are connected by collagen fibres (Fig. 5). Deposition of bone is largely in progress at the edges of the suture, particularly at the tips of the bony serrations (Fig. 9), whereas within the bone both deposition and resorption occur as the normal processes of reconstruction take place.

The teeth and bones are connected by the peridental membrane. At the junction of the membrane and the bone are areas of resorption and deposition of bone and a number of

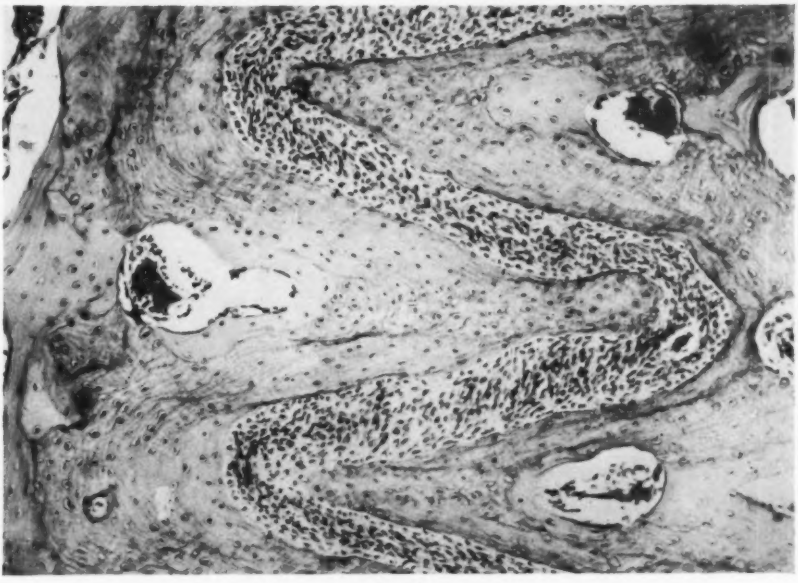

FIG. 5

The normal premaxillary suture in the rabbit, showing long bony serrations held together by a very cellular sutural connective tissue. (×185.)

FIG. 6

The effect of cortisone on the premaxillary suture in the rabbit without mechanical stress. There is increased resorption of bone, with increased vascularity. (×185.)

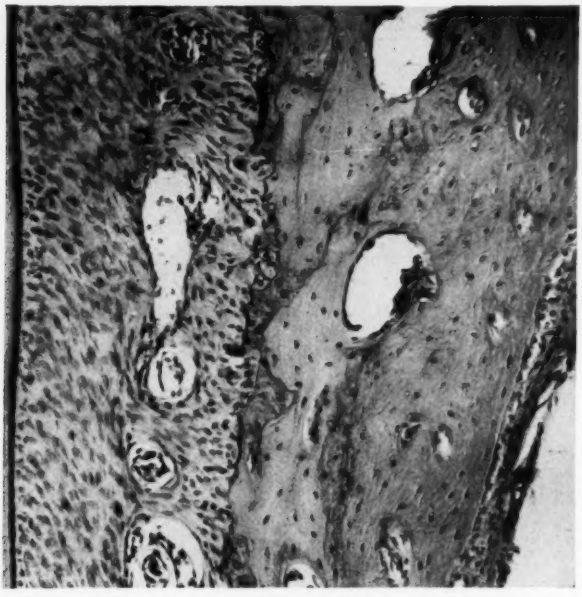

FIG. 7

The normal premaxillary bone and peridental membrane. The edge of the incisor tooth is seen on the left-hand side of the photograph. (×300.)

FIG. 8

The effect of cortisone on the premaxillary bone in the rabbit without mechanical stress. Considerable thinning of bone trabeculae has taken place. (×300.)

FIG. 9

The normal premaxillary suture in the rabbit, without mechanical stress, showing osteoblasts and new bone formation on the tip of one bony serration. (×720.)

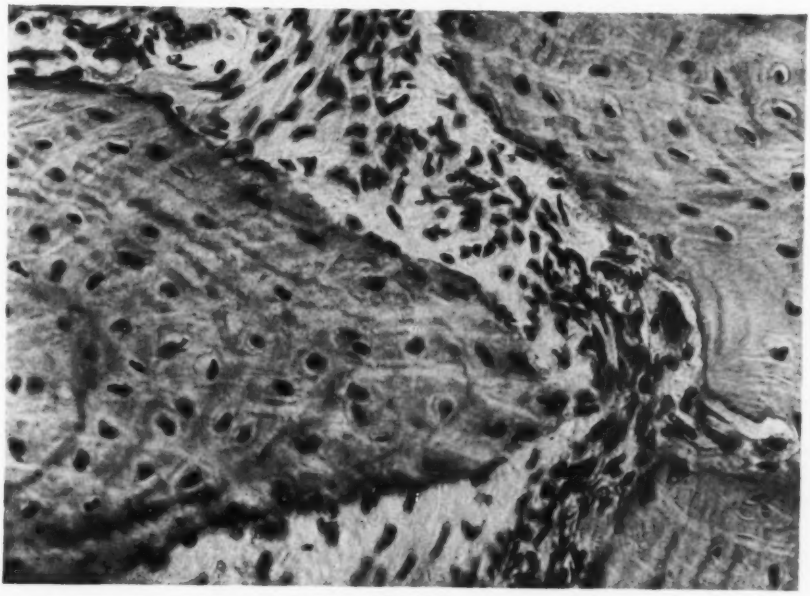

FIG. 10

The effect of cortisone, without mechanical stress, on the premaxillary suture of the rabbit. Osteoblasts are absent on the ends of the bony serration, the bone edges are irregular in outline, and resorption is occurring. In the opposite bone a perforating canal is present, and the fibroblasts of the sutural connective tissue are small, darkly staining and irregularly arranged. (×720.)

haematoxylin-staining reversal lines (Fig. 7). The general arrangement in the normal rabbit is shown in Figure 3.

Cortisone administration—After seven days there is considerable resorption of the bone of the premaxilla. This resorption is seen first (at four days) round marrow spaces and blood vessels and is associated with a great increase in vascularity (dilation of vessels and apparent increase in number) and the presence of small osteoclasts in lacunae (Figs. 6 and 8). Bone deposition, which is normally prominent at this age, is almost absent. This is shown particularly at the ends of the bony serrations of the premaxillary suture, where osteoblasts are absent, and the bone edge is irregular in outline and an osteoid margin is absent (Fig. 10). The sutural connective tissue is also oedematous, and the fibroblasts are smaller and more darkly staining.

By eleven days the bone trabeculae are significantly thinner, although bone resorption is now less obvious. However, under higher magnification, some mononuclear cells, designated as osteoclasts, are still seen associated with continued removal of bone tissue, although the proliferation of connective tissue cells characteristic of many examples of bone resorption (and well exemplified in " osteitis fibrosa ") is not apparent. In some areas cells line the bone where new bone formation is taking place, and these are termed osteoblasts.

The peridental membrane is not obviously changed, though containing some dilated vessels.

A.C.T.H. administration—There was no detectable difference between the animals treated with A.C.T.H. and the normal ones.

Mechanical stress (tooth movement). *Normal rabbit*—When force is applied laterally to the incisor teeth tension is exerted on the bone on one side through the peridental membrane (" tension " side) and pressure is exerted on the bone on the other side (" pressure " side) (Fig. 4).

After four days of tooth movement bone is being laid down on the tension side. The new bone trabeculae are long, thin and orientated in the direction of stress. The longest are near the gingival margin; the trabeculae are progressively smaller towards the apices of the teeth.

Some bone resorption can be seen in the original alveolar bone and in some places the junction of the old and new bone on the peridental membrane side has been penetrated by vascular connective tissue: in some areas osteoclasts are present.

In the peridental membrane an increased number of fibroblasts is orientated in the general direction of the line of stress. The greatest tooth movement has taken place at the gingival margin, and here in the underlying connective tissue is an increase in deeply staining long thin fibroblasts, and there is some intercellular "oedema." Both the staining characteristics and the shape of the fibroblasts change progressively towards the apices of the teeth, where least movement has taken place. A gradation in form from the fibroblast to the osteoblast can be seen near the areas where bone is forming. The osteoblast does not assume the osteocyte form until some time after calcification has begun, and gradations in shape can be seen between the osteoblasts at the site of bone formation and the osteocyte in the first bone laid down.

There is also an increased number of osteoblasts on the ends of the serrations of the suture where the two premaxillary bones are moving apart.

On the pressure side the tooth and bone are almost in contact near the gingival margin, separated by some hyaline material in which the outlines of thin fibroblasts can be detected, orientated at right angles to the line of applied force. Outside this pressure area there is an increase of vascularity in the peridental membrane along its junction with bone, and osteoclasts are present. The blood vessels in the original bone have widened, resulting in further bone resorption, and some new vessels grow through the bone. New bone is forming ahead of the immediate area of pressure on the lateral aspect of the labial plate of bone. This new bone is also orientated in the line of the applied force.

After seven days all these changes have progressed so that the bone trabeculae on the tension sides are larger and thicker (Figs. 11 and 15). The premaxillary bones have moved farther apart and new bone is now being laid down on the edges of the serrations of the suture, particularly near the crest of the alveolar processes (Fig. 13). On the pressure side a large amount of the original bone has been resorbed, and a layer of very cellular bone has formed lateral to it.

On the eleventh day all stages of bone formation and resorption have progressed. Both the new bone trabeculae on the tension side and the serrations of the suture are longer, and considerable resorption of the original bone has taken place.

Cortisone administration—By the end of four days' administration of cortisone there is gross vascularity and extensive resorption of bone within the marrow spaces and around blood vessels within the bone. Sections of premaxillae, progressively farther away from the area of stress, show diminished resorptive changes but, as might be expected, in no case is the normal appearance seen.

Where there is pressure between the moving tooth and the plate of alveolar bone (that is, on the " pressure " side) there is an increased resorption of bone, particularly around vessels within the bone. On the opposite aspect, where the tooth is moving away from the bone and where normally tissue is laid down (that is, on the " tension " side) the appearances at this stage are similar.

Bone formation on the ends of the serrations of the premaxillary sutures is absent, and there is less bone formation at the junction of peridental membrane and bone. There is considerable vascularity of the peridental membrane.

By the seventh day all these changes have progressed; in general the picture is that of intense bone and connective tissue removal, with but little replacement of newly formed bone (Fig. 12). The bony serrations of the suture are being resorbed (Fig. 14), and along the remaining edges of the bone there is absence of osteoblasts (Fig. 18). Instead, the presence of small areas deeply staining with haematoxylin show where small amounts of bone have been resorbed. There is a large increase of vascularity in the sutural connective tissue and, where collagen fibres are inserted into the bone ends they are widely spaced and appear stretched (Fig. 18). There is neither proliferation of connective tissue cells nor their subsequent change to osteoblasts.

On the " tension " side at the junction of bone and peridental membrane new bone trabeculae of normal structure are formed but removed by resorption almost as quickly (Figs. 12 and 16). On the " pressure " side there is gross bone resorption with but little new bone formation. There is extensive vascularity of the peridental membrane, and the fibres become widely separated; there are wide spaces between the collagen fibres, presumably filled with altered (more fluid) ground substance. There is little evidence of repair.

By the eleventh day these changes—vascularity and resorptive processes—have diminished; in some areas osteoblasts are lining bone margins and some bone matrix has formed. At the ends of the bony serrations a layer of homogeneous bone matrix is forming which contains but few osteoblasts and none of the haematoxylin-staining granules characteristic of calcifying bone (Fig. 19). It is separated from the old bone by an irregular darkly staining haematoxylin line which is thicker and more pronounced than those in the surrounding bone. This indicates the site of change from resorption to deposition, or vice versa, and is referred to as the " arrest " or " reversal " line. The collagen fibres of the sutural connective tissue are still widely spaced, even where they enter the newly forming bone matrix.

A similar picture of bone resorption is also seen, both at the junction of bone and peridental membrane and on the pressure side. There are an increase of the ground substance and a separation of the connective fibres of the peridental membrane associated with the continued tooth movement and absence of the cellular activity of repair.

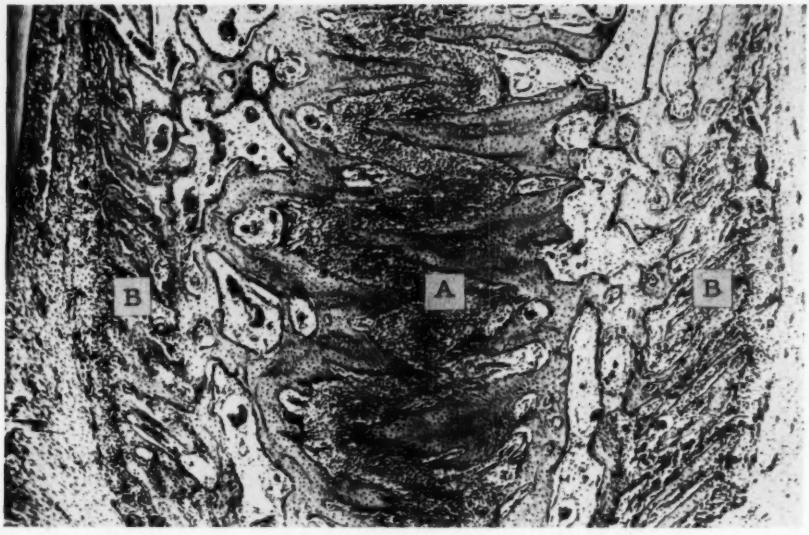

FIG. 11

The premaxillary bones after seven days' tooth movement in a normal rabbit. The bones have moved apart and new bone formation is taking place on the ends of the bony serrations of the suture where they are under tension (A). As the incisor teeth move apart tension is also exerted by the connective tissue of the peridental membrane at the junction of the peridental membrane and premaxillary bones. New bone trabeculae, which are orientated in the direction of the applied tension (B), form here. (×40.)

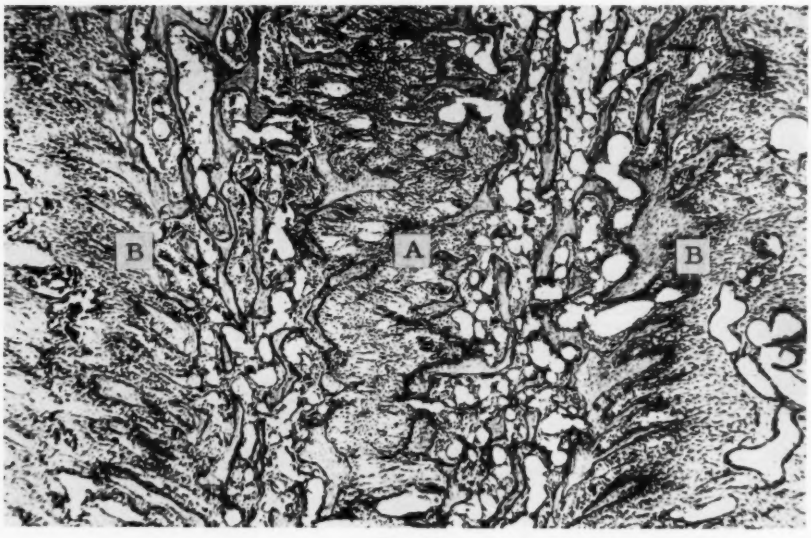

FIG. 12

The effect of cortisone on the premaxillary bones after seven days' tooth movement in the rabbit. Instead of new bone formation there is extensive resorption of bone and increased vascularity; small haemorrhages are present. There is complete failure of new bone formation on the ends of serrations of the premaxillary suture (A) and partial failure of new trabecular formation from the junction of the peridental membrane and premaxillary bone (B). (×40.)

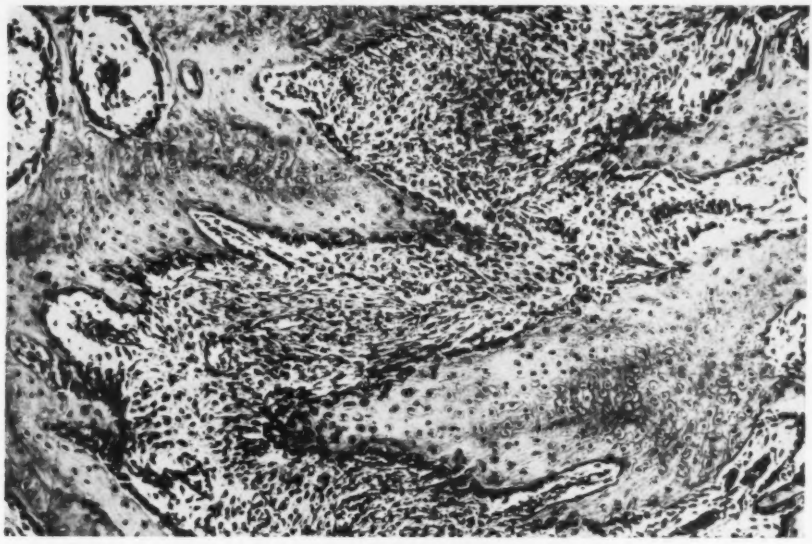

FIG. 13

The serrations of the premaxillary suture after seven days' tooth movement in a normal rabbit. New bone formation is taking place on the ends and edges of the serrations where tension is being applied. This is shown by the orientation of the sutural connective tissue. (×185.)

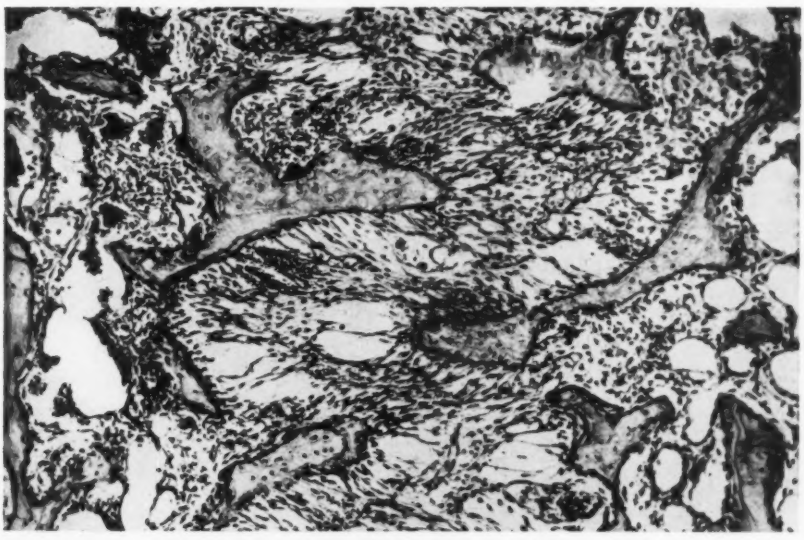

FIG. 14

The effect of cortisone on bony serrations of the premaxillary suture after seven days' tooth movement in the rabbit. Extensive resorption of bone has taken place so that the ends of the serrations are almost separated from the body of the premaxillary bones. This resorption is associated with a great increase of vascularity and appearance of osteoclasts at bone edges. There is but little new bone formation occurring at the ends and edges of the bony serrations. The fibres of sutural connective tissue are widely separated and fibroblasts are smaller than in the normal animal. (×185.)

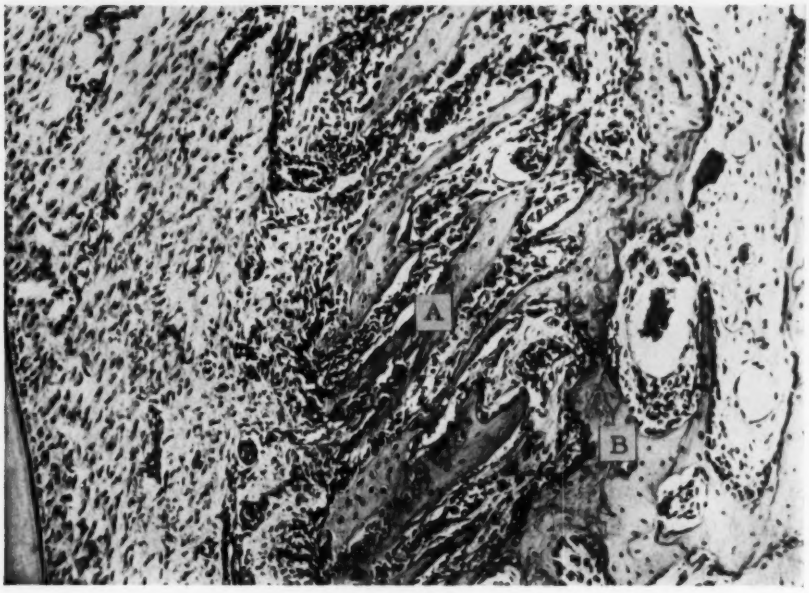

FIG. 15

The formation of new bone trabeculae (A), orientated in the direction of tension applied by the movement of incisor teeth for seven days. These new bone trabeculae arise from the junction of the premaxillary bone (B) and the peridental membrane. (×185.)

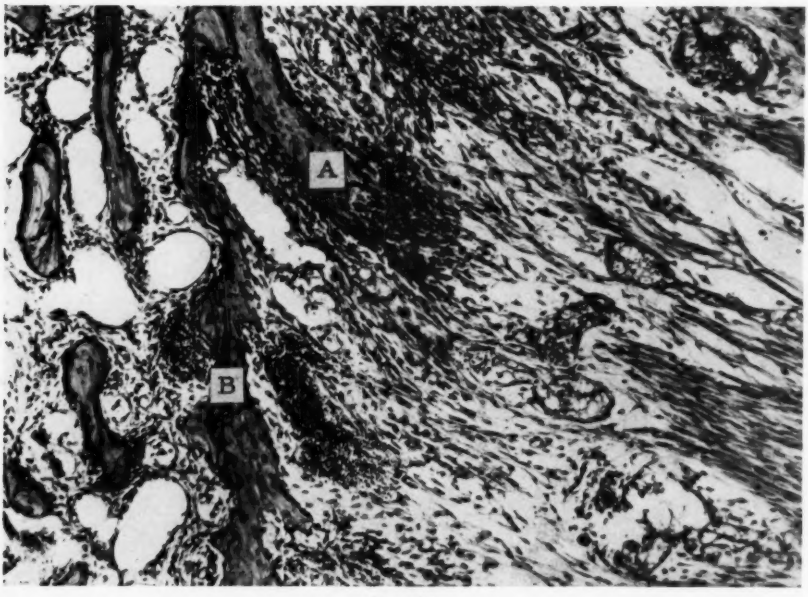

FIG. 16

The effect of cortisone on the formation of new bone trabeculae (A) after seven days' tooth movement. Only a few new bone trabeculae have formed; there is an increased oedema and vascularity of connective tissues with some areas of haemorrhage present. Osteoclasts are present at the bone edges and the premaxillary bone (B) is being resorbed rapidly. (×185.)

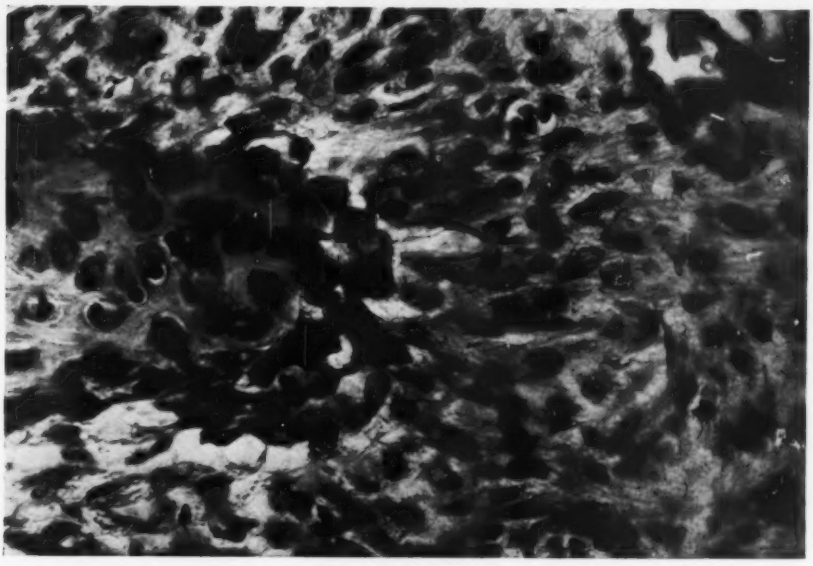

FIG. 17

The tip of a bony serration of the premaxillary suture after seven days' tooth movement in the normal rabbit. New bone is being formed; large numbers of fibroblasts from the sutural connective tissue are transformed into osteoblasts and new bone matrix is already forming. (×720.)

FIG. 18

The effect of cortisone on the tip of a bony serration of the premaxillary suture after seven days' tooth movement in the rabbit. There is absence of proliferating fibroblasts, the cells present are small and darkly staining and osteoblasts are not present at the bone edge. The connective tissue fibres are widely separated and orientated in the direction of tension. Resorption of bone has separated this tip of a serration from the adjoining bone. (×720.)

A.C.T.H. administration—There is no detectable change in mechanically stressed bone and connective tissue when A.C.T.H. is administered to the rabbit; experiments with this hormone were extended to study the effect of treatment with A.C.T.H. before tooth movement. It was administered for seven and fourteen days before tooth movement was begun and continued for its duration. There is a significant difference from normal after twenty-one days'

FIG. 19

The effect of cortisone on the tip of a bony serration of the premaxillary suture after eleven days' tooth movement in the rabbit. Some new bone matrix has been laid down on the tip of the serration; a deeply staining " reversal " line separates the old and new bone. The new bone contains fewer osteoblasts compared with the normal animal after eleven days' tooth movement. The nuclei of the fibroblasts of the sutural connective tissue are darkly staining and spindle shaped; few osteoblasts are present at the edge where new bone is forming. (×720.)

administration of A.C.T.H. and seven days' tooth movement. Bone resorption and vascularity in the suture of the premaxilla are slightly increased, but less than in the animal treated with cortisone.

Sections of the lower end of the femur were made to see whether bone growth was affected. After fourteen and twenty-one days' administration growth of the epiphysial cartilage was inhibited (Storey 1957).

THE GUINEA PIG

No mechanical stress. *Normal guinea pig*—In the normal guinea pig, aged eight to twelve weeks, two premaxillae have united and all signs of a suture are obliterated.

Cortisone administration—There was no demonstrable effect of cortisone (5 milligrams a day) on the normal premaxillary structures.

A.C.T.H. administration—There was no demonstrable effect on bone or connective tissue.

Mechanical stress (tooth movement). *Normal animal*—This has been described in detail before (Storey 1955*b*). The changes resemble those in the rabbit except that the premaxillary bones do not separate.

After seven days' tooth movement the trabeculae on the tension side are long and arranged along the line of stress, as in the rabbit. Some resorption of bone on the pressure and tension sides has also taken place.

Cortisone administration—Five milligrams a day of cortisone produced no demonstrable change from the normal pattern of bone change associated with tooth movement.

With the higher dose, 10 milligrams a day, there is slight retardation of bone formation on the tension side at the junction of bone and peridental membrane. This is seen first at three days. Bone resorption is not accelerated on either the " tension " or " pressure " sides.

A.C.T.H. administration—New bone formation is slightly delayed in areas under greatest tension—that is, at the alveolar margin on the " tension " side. Here, where the collagen fibres of the peridental membrane are running through the new bone, swollen osteoblasts are present between the fibres, but bone matrix is not seen. There is no increased bone resorption on either " pressure " or " tension " sides.

DISCUSSION

The most significant finding in these experiments is that the administration of cortisone in the rabbit is followed by rapid resorption of bone in both normal conditions and under mechanical stress. The observations made on the guinea pig emphasise the importance of recognising species differences. Where resorption occurs under stress—for example, on the pressure side of a moving tooth—resorption is increased. The early bone resorption seen after seven days' cortisone administration is characterised by an increase of vascularity, and the numerous small cells lining the bone surfaces are clearly osteoclastic in function. Bone formation occurs during tooth movement on the " tension " side, but after injections of cortisone it is either absent, or where new trabeculae form they are resorbed almost as rapidly as they are laid down. These changes are different from those in the normal animal, where the formation of new bone and connective tissue maintains the continuity of the remodelling tissues associated with the tooth movement.

In later stages of the experiment there is a reduction of the initial resorptive processes and some poorly formed bone is seen on the ends of the serrations of the sutures. A haematoxylin-staining line separates the old and new bone, which is thicker and darker than that seen in normal bone. The thick lines probably indicate both the delay in formation of bone matrix and the secretion of abnormal amounts or kinds of cementing substances. In the early stages of resorption the spaces left by the removal of bony structure are filled with dilated blood vessels, some poorly demarcated strands of eosinophilic-staining material and small fibroblasts. By eleven days this change is subsiding and resorption spaces in continuity with bone marrow are being filled by marrow cells. This absence of a " fibrous tissue " replacement of bone is consistent with the anti-anabolic (Albright 1943) and antimitotic (Green and Ghadially 1951) effects of cortisone on tissues.

The rapid bone resorption caused by cortisone has not been recorded before. Sissons and Hadfield (1951) found that cortisone inhibited the healing of fractures of bones in the rabbit, as had Blunt and his associates (1950). Follis (1951*a*) found that cortisone interferes with the normal osteolytic sequences in the rat and produces changes at the epiphysial cartilage like those seen after administration of oestrogen, whereas Key, Odell and Taylor (1952) failed to show any effect of cortisone on the healing of fractures in rats. Later Sissons and Hadfield (1955) showed that bone changes like those in Cushing's syndrome can be produced in rabbits' femurs by the administration of cortisone. Sissons (1956) attributed the loss of bone substance to inhibition of bone growth while normal osteolytic processes continue.

Follis (1951*b*) commented on the absence of osteoclasts in bones in Cushing's syndrome and found it difficult to account for the loss of bone substance from a study of histological sections. Bone resorption is a physico-chemical event which may be mediated, in part, by some specific cells. Commonly, the bone cells influence the formation-resorption process in either direction. The formation of special cells, to be designated osteoclasts, is an incidental feature which is found in only some circumstances. The absence of this inconstant morphological component in any particular circumstance does not affect the associated observations.

In the rabbit these cells are a prominent feature in the early stages of the action of cortisone; they decrease in number with time, even though bone removal continues. It is apparent from the different histological pictures presented that rapid bone resorption can take place in one of two ways: 1) with replacement of fibrous connective tissue (fibrous type of reaction); and 2) with no replacement by connective tissues, but instead marrow cells and fat eventually filling the spaces left by the removal of bone.

The structural arrangement of stressed connective tissues in the cortisone-treated rabbit is also striking. Collagen fibres are no longer held together as they are normally, but are stretched and widely separated, indicating a change in the physical properties of the ground substance. Such a physical (and chemical) change would naturally alter the mechanical properties of ground substance of bone and would be expected to lead to increased bone resorption from the action of normal stresses alone. At the same time the anti-anabolic effect of cortisone would inhibit the replacement or addition of bone and would thus cause extreme thinning of existing bone trabeculae.

The failure of A.C.T.H. to produce any significant change may possibly be correlated with the observation that the rabbit secretes relatively little hydrocortisone (Bush 1953). Differences between cortisone and A.C.T.H. have been found in skin-grafting experiments in the rabbit, where A.C.T.H. failed to affect the tissues in the same way as cortisone (Krohn 1954). The slight change in the effect of A.C.T.H. on tissues during tooth movement after three weeks' administration is consistent with Kass, Hechter, Macchi and Mou's (1954) observation that the ratio of hydrocortisone to corticosterone secreted by the adrenal gland changes so that after three to four weeks' stimulation with A.C.T.H. there was a large increase in hydrocortisone.

There are known species differences in the effect of cortisone on tissue reactions: the rabbit is one of the most sensitive and the guinea pig one of the least sensitive (Sparrow 1953). The findings in this paper are consistent with these known species differences.

Cortisone leads to porosis in man also, either during its therapeutic administration (Boland 1951, *British Medical Journal* 1955) or in Cushing's syndrome. This suggests that the results of rabbit experiments are applicable in man, particularly in showing that, in areas of bone under abnormal stresses, rapid resorption of bone may be expected to take place.

SUMMARY

1. When cortisone is administered to rabbits there is early rapid resorption of bone and a partial inhibition of new bone formation. After a few days the effect becomes less obvious, so that, if observations are made at later stages, the results may be ascribed then to simple inhibition of bone growth.

2. The effect of mechanical stress has been studied in the jaw. When tooth movement is induced mechanically there is, in ordinary circumstances, a resorption of bone on the side to which the tooth is moving (the " pressure " side) and bone formation on the opposite side (the " tension " side). After administration of cortisone there is increased resorption on the " pressure " side and there is greater resorption of connective tissues here. On the " tension " side there is resorption and inhibition of bone formation.

3. In the areas of stress, when cortisone is administered, collagen fibres are no longer in apposition, being separated by spaces presumably filled with altered ground substance; this kind of change may be responsible for many of the observed phenomena.
4. A.C.T.H. does not produce a demonstrable resorptive effect on bone or connective tissue until it has been administered for periods longer than is required for cortisone (three weeks); even then the change is not pronounced.
5. In the guinea pig there is slight delay in bone formation with large doses of both cortisone and A.C.T.H., but no significant bone resorption occurs.

Thanks are due to Messrs Merck and Co. Inc., who generously supplied the cortisone acetate used in these experiments, and to the Commonwealth Serum Laboratories, Australia, who provided the A.C.T.H.

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PRIMARY BONE-FORMING TUMOURS AND THEIR RELATIONSHIP TO SKELETAL GROWTH

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The frequency with which the relatively rare tumours of bone arise in young persons is well known. Kolodny (1927) has fully described the bone sarcomata, and the Bone Sarcoma Register of the American College of Surgeons has given rise to an extensive literature. The benign bone-forming growths number two, namely osteochondroma (exostosis) and osteoma (or fibro-osteoma). Three large series of osteochondromata have been reported—by Stocks and Barrington (1925), by Meyerding (1927) and by Geschickter and Copeland (1949).

From all these records the age, sex and site distribution have been broadly outlined. The present communication presents an analysis of groups of primary bone-forming tumours with special reference to the juvenile period of life, and how their occurrence is related to skeletal growth.

There is still some confusion in the nomenclature of bone tumours; thus the scope of this paper must first be defined.

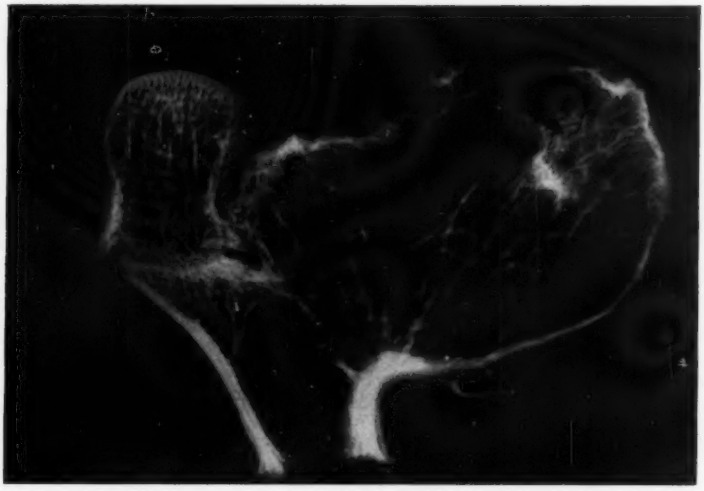

FIG. 1

Osteochondroma of metatarsal from boy aged fourteen. Radiograph (×2) of a longitudinal slice of the surgical specimen. Note relationship of the tumour to the epiphysial growth cartilage, and the absence of metaphysial cortex in the base of the tumour.

TUMOURS UNDER CONSIDERATION

Benign bone-forming tumours. *Osteochondroma*—This tumour may be multiple (Keith's aclasis) or solitary, the two forms having the same essential structure. The former is best regarded as a genetically conditioned connective-tissue dystrophy characterised by the appearance and growth of numerous osteochondromata during skeletal development. Such cases have been

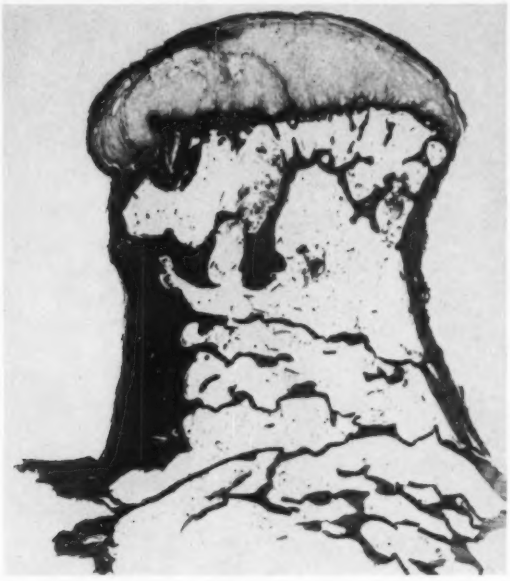

FIG. 2

Osteochondroma of femur from man aged twenty-three. Photomicrograph of the complete tumour. (Haematoxylin, phloxin and tartrazine, ×5.) Note the zone of ossification in the deeper part of the chondroid cap; also the formation of a "distinct shaft" on which new periosteal bone is laid down to form, in effect, a supernumerary tubular bone.

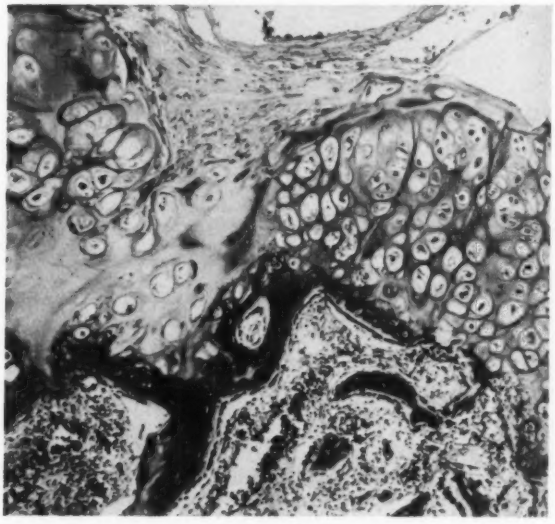

FIG. 3

Osteochondroma of femur from a boy of fifteen. Photomicrograph. (Haematoxylin, phloxin and tartrazine, ×60.) The growth cap, showing the distorted manner in which the structure of the normal growth cartilage is reproduced.

excluded from the present study because documentation is seldom adequate. Furthermore, Stocks and Barrington (1925) have shown that the hereditary element has a markedly male predominance, and influences both site and age of the appearance of the multiple tumours.

FIG. 4

Osteogenic sarcoma of humerus (chondroblastic type) from a boy aged eleven. Radiograph (×1) of a longitudinal slice of the complete tumour. This sarcoma was of histological Grade II. The soft-tissue mass, which extends equally medially and laterally, consisted of chondrosteoid tissue (Fig. 6) intermingled with cellular areas (Fig. 7). The deeper parts of this aggressive tumour were fairly widely ossified, especially towards the lower part, where a well defined "Codman's triangle" can be seen.

The solitary osteochondroma may be defined as an accessory ossicle, which grows through the intermediate stage of a chondroid cap which mimics the structure and behaviour of a normal growth cartilage (Fig. 3).

For the purposes of the present analysis, the most valuable series of these tumours were those fully reported by Geschickter and Copeland (1949). The Bristol Bone Tumour Register contains but thirty-one examples. These, with a few others, have served for radiographic and histological study, but are too few for statistical analysis (Figs. 1 to 3). " Osteochondroma " must be distinguished from " ossified chondroma." This, usually centrally placed in bone, consists of small-celled cartilage in whose matrix calcification is common, but true ossification infrequent and atypical.

Osteoma—This innocent tumour arises mainly from the membrane bones of the skull, and is rare. Unlike the osteochondroma, it is ossified from condensed fibrous tissue, cartilage being absent. It is not mentioned further because the present paper is concerned only with tumours related to the growth in length of bones, which is entirely endochondral.

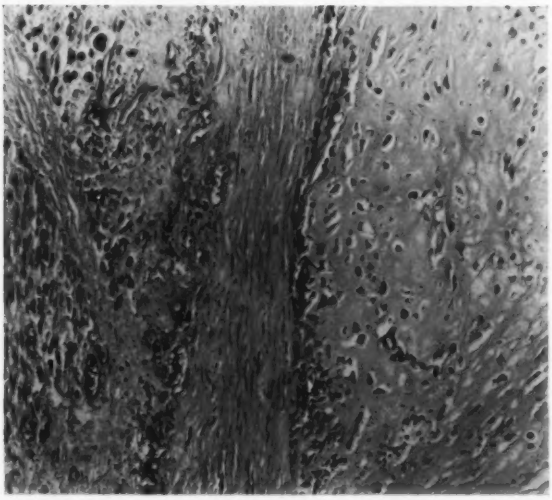

FIG. 5

Osteogenic sarcoma (same case as in Figure 4). Photomicrograph. (Haematoxylin, phloxin and tartrazine, × 105.) The advancing edge of the tumour, showing the proliferating cambial layer. There is also much interstitial cell growth. (Condensed fibrous tissue and atrophic muscle to the left.)

Malignant bone-forming tumours. *Osteogenic sarcoma* (Figs. 4 to 8)—This name is retained for a sarcoma that arises from bone—in the sense originally used by Ewing (1922). Two other names are often used synonymously: " osteosarcoma " and " osteoblast sarcoma." The former name is acceptable, but the latter rests on no certain foundation, as in many tumours the identity of the atypical and pleomorphic cells with osteoblasts is presumptive and debatable.

Primary chondrosarcoma—This is a variant of osteogenic sarcoma in which most of the tumour tissue is neoplastic cartilage. It is usually subperiosteal and often nodular, the individual nodules of tumour tissue showing evidence of both appositional and interstitial growth. Bone formation is mainly endochondral; and many of the cells, although atypical, may still be recognised as chondroblasts or chondrocytes (Figs. 6 and 7). It is not uncommon, however, to find areas in which the sheet of matrix is broken up into a trabecular pattern, with deposition of osteoid by tumour cells more closely resembling osteoblasts. This tumour is said to be less aggressive than the primarily osteoid-forming types of osteogenic sarcoma, although Thomson and Turner-Warwick (1955) have shown certain specimens to be very active in their growth

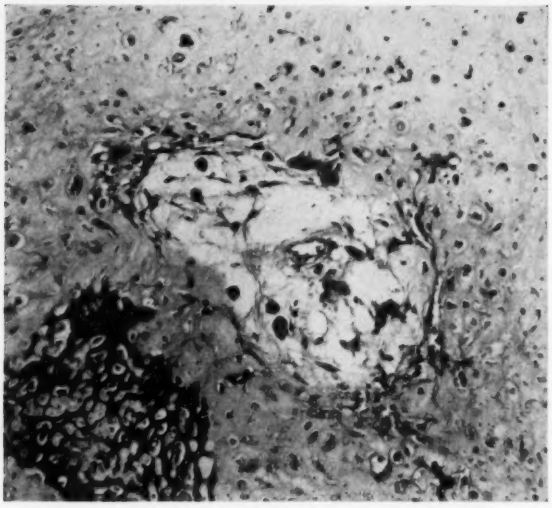

FIG. 6

Osteogenic sarcoma (same case as in Figure 4). Photomicrograph. (Haematoxylin, phloxin and tartrazine, ×105.) Field of the deep tumour parenchyma showing the relatively acellular nature of the chondrosteoid matrix. There were numerous foci of active resorption by osteoclasts, many of which were of atypical appearance.

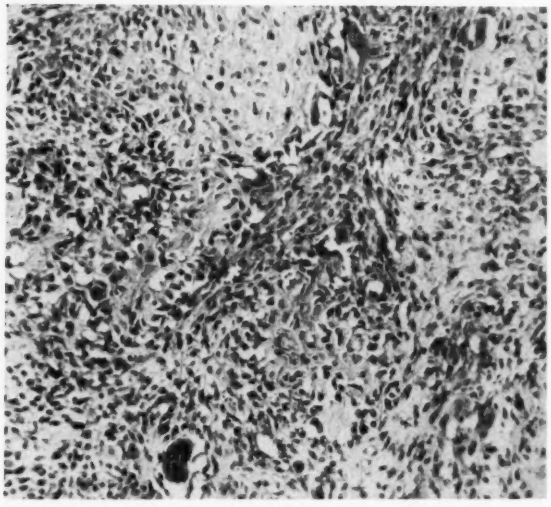

FIG. 7

Osteogenic sarcoma (same case as in Figure 4). Photomicrograph. (Haematoxylin, phloxin and tartrazine, ×105.) Typical field of cellular tumour tissue, showing atypism and pleomorphism of cells with formation of tumour osteoid. It is these tumour cells that may defy definite decision as to their nature—osteoblast or chondroblast. Bizarre giant-cell forms are common.

and spread. The separation of this tumour as an entity may be justified upon clinical grounds; nevertheless, many such tumours form bone, sometimes extensively. I prefer the term " chondroblastic osteogenic sarcoma " for those personally studied. Seven such neoplasms from the Bristol Bone Tumour Register are included in the present group.

FIG. 8

Osteogenic sarcoma of fibula from a woman aged thirty-four. Radiograph (×1·5) of longitudinal slice of complete tumour. This sarcoma was of histological Grade I. Much of the soft-tissue shadow consisted of chondrosteoid matrix. In view of the slower growth of this tumour, new bone formation is well shown and relatively regular (*cf.* Fig. 4). The tumour cells were mainly of osteoblast type.

AGE INCIDENCE

Figure 9 and Table I show the data for relative tumour incidence (corrected for population distribution) of two series of tumours: 240 osteochondromata (Geschickter and Copeland 1949); and 140 osteogenic sarcomata (Bristol Bone Tumour Register series of sixty-seven

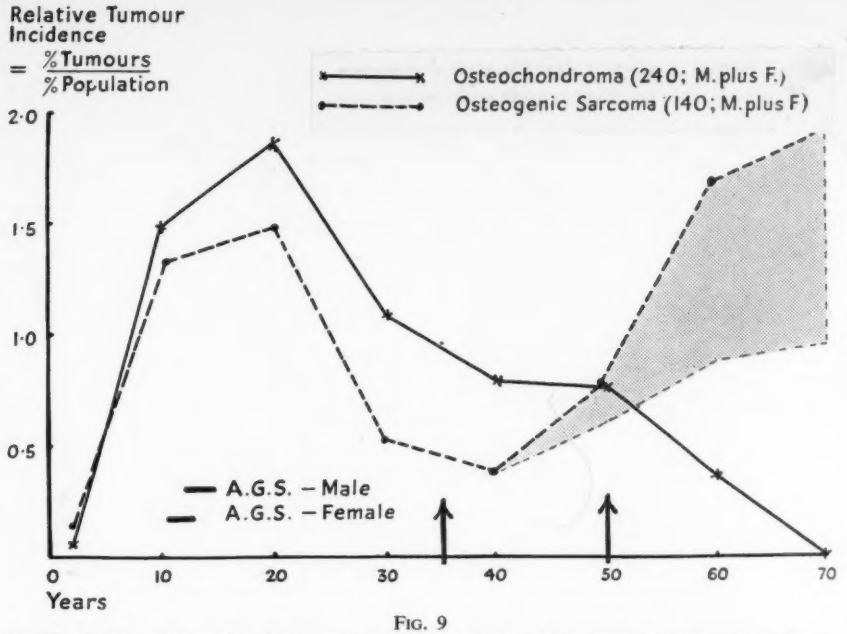

FIG. 9

Relative incidence of bone-forming tumours at various ages. Data from Table I. The vertical pointers indicate subdivision of the tumours by age into three groups—juvenile, intermediate, senile. The shaded area indicates the observed proportion of sarcoma cases associated with Paget's osteitis deformans. A.G.S. = Adolescent growth spurt.

tumours, plus seventy-three others reported in the British Empire Cancer Campaign Annual Report (1949)). The Bristol series represents all tumours registered during 1946–52, and probably includes the great majority of all such growths which appeared in persons dwelling in the south-west region of Great Britain at that time. By comparison, the other series of seventy-three sarcomata was collected from many sources, and is hence probably a random sample also devoid of any institutional bias.

TABLE I

	Age in years									Total
	0–4	5–	15–	25–	35–	45–	55–	65–	75–	
Osteochondroma (M+F)										
Number of tumours .	1	60	80	43	27	22	7	—	—	240
Relative tumour incidence	0·05	1·47	1·85	1·09	0·79	0·76	0·36	—	—	—
Osteogenic sarcoma (M+F)										
Number of tumours .	1	24	27	11	8	15	25	20	9	140
Relative tumour incidence	0·13	1·31	1·49	0·51	0·38	0·77	1·68	1·91	1·60	—

Note—The relative tumour incidence is calculated by separating the whole series by age as shown above, and dividing the percentage of tumours for each group by the appropriate percentage of population. Each figure is relative only to its own series. To compare actual frequencies in two such series it is necessary to reduce both to the same total number of tumours (see Fig. 6).

The curves depicted in Figure 9 have been considered elsewhere (Price 1955), and I will consider here, in rather more detail, the data which apply only to persons under thirty-five years of age. Growth and maturation of the skeleton continue into the third decade, to be accompanied by a few typical tumours (both innocent and malignant) in the middle and late twenties. In order, however, to apply correction factors for population-age distribution, it is necessary to use statistical tables in which the subdivision by age is 0–4 years, 5–14 years,

TABLE II

Tumour	Number	Mean age (*years*)	Males		Females		Difference in mean age M—F (*years*)	Standard error of difference
			Number	Mean age (*years*)	Number	Mean age (*years*)		
Osteochondroma .	136	15·9	95	16·4	41	15·0	1·4	1·30
Osteogenic sarcoma	36	16·8	25	17·2	11	16·0	1·2	1·89

Note—Age of patients all taken at time of presenting symptom.

TABLE III

OSTEOGENIC SARCOMA

Site	Total number	Mean age (*years*)	Males		Females		Difference in mean age M—F (*years*)	Standard error of difference
			Number	Mean age (*years*)	Number	Mean age (*years*)		
Femur .	19	17·3	11	17·0	8	17·7	−0·7	2·51
Tibia .	7	16·7	6	17·8	1	10·0	7·8	—
Humerus	10	15·9	8	17·0	2	11·5	5·5	3·16

TABLE IV

OSTEOGENIC SARCOMA

Site	Total number	Mean age (*years*)	Males		Females		Difference in mean age M—F (*years*)	Standard error of difference
			Number	Mean age (*years*)	Number	Mean age (*years*)		
Femur .	40	15·7	25	16·1	15	14·9	1·2	1·54
Tibia .	28	15·8	17	15·9	11	15·7	0·2	1·19
Humerus	16	13·3	5	17·2	11	11·6	5·6	2·14
F+T+H	84	15·3	47	16·1	37	14·2	1·9	0·94

15–24 years, and thence by decades. Consequently, the juvenile period here under review has been taken to extend from birth until thirty-five years of age. Table II presents an age and sex analysis of all tumours of both series included in Table I, occurring within the group 0–35 years, and in which the sex of the patient was recorded.

From Figure 9 and the data in Tables I and II the following trends can be discerned. 1) There is close similarity in the juvenile portions of the age incidence curves of these two

groups of osteochondromata and osteogenic sarcomata. The former curve, however, continues to fall steadily during life, whereas the latter rises again during the fifth decade to reach its zenith during the sixth and seventh decades. This rise is largely due to Paget's osteitis deformans, which is associated with at least 50 per cent of osteogenic sarcomata after fifty years of age (Price 1955, see also Figure 9). 2) In both tumours the mean age for females tends to be less than that for males by about the same amount (twelve to fourteen months).

Osteogenic sarcoma—Although the Bristol Bone Tumour Register's group of juvenile osteogenic sarcomata included in Table II is small, the difference between mean age in the two sexes may also appear when the series is broken down into groups of tumours arising in separate bones. This is shown in Table III.

TABLE V

OSTEOGENIC SARCOMA

(See also Figure 7)

	Age (*years*)				Total	Group mean age (*years*)	Total mean age (*years*)
	0–4	5–	15–	25–			
Males							
Femur	—	15	17	4	36	16·4	16·6
Tibia	—	5	18	—	23	16·3	
Humerus	—	5	7	1	13	17·1	
Totals	—	25	42	5	72		
% Tumours	—	34·6%	57%	8·4%	100%		
% Population	13·8%	29·1%	30·3%	26·8%	100%		
Relative tumour incidence = $\frac{\%T}{\%P}$	—	1·2	**1·88**	0·3	—		
Females							
Femur	—	9	12	2	23	15·9	14·5
Tibia	—	5	7	—	12	15·3	
Humerus	—	10	3	—	13	11·5	
Totals	—	24	22	2	48		
% Tumours	—	50%	45·7%	4·3%	100%		
% Population	12·9%	28·2%	30·7%	28·2%	100%		
Relative tumour incidence = $\frac{\%T}{\%P}$	—	**1·8**	1·5	0·2	—		

Note—The data presented in this table (and Table III) are not significantly modified by a correction calculated for young men absent from the Bristol B.T.R. series on active service.

The data given in Table III may be compared with figures similarly derived from a larger group of eighty-four osteogenic sarcomata in persons under thirty-five years of age culled from the tumours listed by Geschickter and Copeland (1949).

From these tables the following trends may be noted. 1) Bone for bone, the mean age is usually less in the female than the male. 2) The mean age (in both sexes) tends to diminish for the long bones in the following order: femur, tibia, humerus, although, with the modest number of cases in each group the two former bones may not always clearly show this feature. The figures for the humerus are undoubtedly weighted by the relatively large number of females included (50 per cent of the combined totals in Tables III and IV).

The two series of osteogenic sarcomata analysed in Tables III and IV have been combined, as there is no reason to suppose that any extraneous factor has influenced the selection and

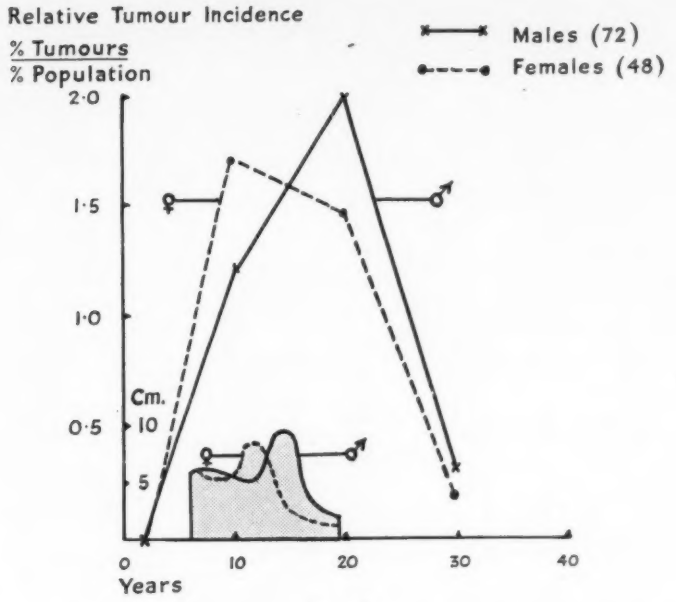

FIG. 10

Relative incidence of osteogenic sarcoma in males and females at various ages up to thirty-five years. (Data for annual gain in height drawn from Tanner (1955).)

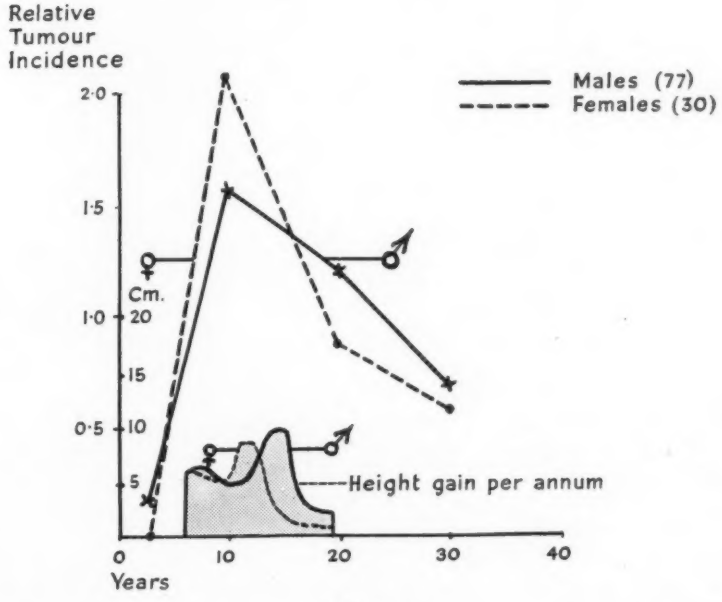

FIG. 11

Relative incidence of osteochondroma in males and females at various ages up to thirty-five years. (Data for annual gain in height drawn from Tanner (1955).)

recording of these tumours. This total group of 120 sarcomata (all in persons under thirty-five years of age) has been further studied for site, age and sex distribution with the results given in Table V and depicted in Figure 10. In this diagram the curves for annual gain in height have also been shown.

From Table V may be noted the relatively larger proportion of these sarcomata of long bones which arise in girls from age five to fourteen years—50 per cent of the total, as compared with 34·6 per cent in males.

Osteochondroma—The general trend of differences in mean age between the sexes which has been indicated for osteogenic sarcoma (Tables II, III and IV) appears also in the analysis of 136 osteochondromata in persons under the age of thirty-five years. These tumours were again selected from Geschickter and Copeland's Tables 2 and 3, and form part of the larger series of osteochondromata of all ages that have been analysed for age incidence in Table I.

None of the differences in mean age in the above table is statistically significant. It is, however, of interest that the differences here, as with osteogenic sarcoma (Tables II, III and IV),

TABLE VI

OSTEOCHONDROMA

Site	Total number	Total per cent	Total mean age (*years*)	Males		Females		Difference in mean age M—F (*years*)	Standard error of difference
				Number	Mean age (*years*)	Number	Mean age (*years*)		
Femur . .	51	37·5	17·4	35	17·5	16	17·1	0·4	2·26
Tibia . .	26	19·1	13·8	20	13·9	6	13·3	0·6	2·04
Humerus .	30	22·1	13·3	22	14·0	8	11·4	2·6	3·30
Pelvis . .	7	5·2	20·4	5	20·6	2	20·0	0·6	—
Vertebra .	4	2·9	10·3	3	11·7	1	6·0	5·7	—
Scapula .	5	3·7	10·6	2	9·5	3	11·3	−1·8	—
Tubular bones	13	9·5	20·7	8	22·6	5	17·6	5·0	—

Note—The groups of tumours of the three major long bones included in Table VI have also been analysed for age and sex distribution, the data for relative tumour incidence being shown in Table VII and Figure 8.

are predominantly in one direction; that is, the mean age tends to be less in the female than in the male. It should, however, be pointed out that in the larger groups of cases, where the figures are presumably more reliable, this difference in mean age is of the same order as the greatest difference in bone age between the sexes during skeletal growth. It may further be emphasised that the firm foundation for the physiological sex dimorphism of bone growth rests mainly upon " longitudinal " studies of large numbers of children. It would be virtually impossible to apply this anthropometric method to children with tumours. Thus any improvement in the statistical value of the data in the foregoing tables can be obtained only by the close consideration of much larger numbers of fully documented cases of these two new growths.

Table VI shows that the relationship of site to age resembles that already noted for osteogenic sarcoma. That is to say that the mean age at which the tumours arise tends to diminish for the long bones in the following order: femur, tibia, humerus. It seems from Table VII that this feature is common to both sexes, and in the analysis of age/sex/site in Table VII it may be noted that during the second decade a larger proportion of osteochondromata appeared in the tibia and humerus than in the femur.

TABLE VII

OSTEOCHONDROMA

(See Figure 8)

	Age (*years*)				Total	Bone group mean age (*years*)	Total mean age (*years*)
	0–4	5–	15–	25–			
Males							
Femur	—	11	17	7	35	17·5	15·8
Tibia	—	12	5	3	20	13·9	
Humerus	2	12	4	4	22	14·0	
Totals	2	35	26	14	77		
% Tumours	2·8%	45·5%	33·6%	18·1%	100%		
% Population	13·8%	29·1%	30·3%	26·8%	100%		
Relative tumour incidence = %T/%P	0·23	**1·56**	1·1	0·67	—		
Females							
Femur	—	8	3	5	16	17·1	14·8
Tibia	—	4	2	—	6	13·3	
Humerus	—	6	2	—	8	11·4	
Totals	—	18	7	5	30		
% Tumours	—	60%	23%	17%	100%		
% Population	12.9%	28·2%	30·7%	28·2%	100%		
Relative tumour incidence = %T/%P	—	**2·12**	0·75	0·60	—		

Note—It may be seen from Table VII that on treating 107 osteochondromata in persons under the age of thirty-five years as a separate series, the highest figure for relative tumour incidence obtains in females of the age period five to fourteen years, i.e., a relatively larger number of these tumours appear in females at an earlier age than in the male. (The tumours which have been separated for age, sex and site are those listed in the like order in Table VI.)

In this smaller series at a restricted age, the humerus is second in frequency; however, in the much larger group of osteochondromata *of all ages* analysed in Table VIII the usual order of bone distribution is shown.

An attempt was made to obtain further information about the mean ages of appearance of osteochondromata of the ilium and scapula, because the few cases included in Table VI showed a marked difference in these two flat bones. A report by Du Mortier and Halpert (1934) included twenty such tumours of the scapula (eighteen culled from the literature), and others were collected, establishing a group of thirty-three osteochondromata of scapula, twenty-nine in persons under thirty-five years of age. Derived data from these are given below.

Total number	Mean age	Males		Females	
		Number	Mean age	Number	Mean age
29	9·8 years	16	9·7 years	13	9·9 years

The relatively early age at which this tumour arises may be established from this larger group; but there is virtually no difference in the mean age at which the tumour appears in the sexes. (This may possibly be related to the characteristically greater development of the male shoulder girdle.)

By comparison, the mean age of a series of forty osteochondromata of the pelvis reported by Ghormley *et al.* (1946) was found to be thirty years. The details given of some are insufficient for a distinction between osteochondroma and ossifying chondroma, and the patient's sex was often omitted. Nevertheless, this material certainly suggests that pelvic tumours arise mostly a few years later than those of the scapula. *Per contra*, the site of the scapular tumour would favour earlier clinical recognition.

TABLE VIII

OSTEOCHONDROMA

Site	Type of bone	Number	Per cent	Combined per cent
Femur	Long	263	26·9	61·4
Tibia	Long	150	15·5	
Humerus	Long	123	12·6	
Fibula	Long	36	3·7	
Radius	Long	15	1·5	
Ulna	Long	12	1·2	
Phalanx	Tubular, short	30	3·1	10·3
Metacarpal	Tubular, short	2	0·2	
Metatarsal	Tubular, short	3	0·3	
Hand*	Tubular, short	14	1·4	
Foot*	Tubular, short	52	5·3	
Clavicle	Long, membrane	29	3·0	3·0
Vertebra	Irregular	30	3·1	6·8
Os calcis	Irregular	28	2·9	
Carpal and tarsal	Irregular	8	0·8	
Patella	Sesamoid, round	5	0·5	0·5
Scapula	Flat	58	5·9	14·2
Ilium	Flat	18	1·8	
Ischium	Flat	4	0·4	
Pubis	Flat	8	0·8	
Pelvis*	Flat	45	4·6	
Sternum	Flat	1	0·1	
Ribs.	Flat	6	0·6	
Skull*	Membrane	38	3·8	3·8

* Precise site not stated.

ANATOMICAL DISTRIBUTION

Osteochondroma—Table VIII gives the distribution according to site of 978 osteochondromata *of all ages*. (The data represent the combination of the three large series formerly mentioned. Each series separately shows the diminishing frequency of involvement of the long bones in the following order: femur, tibia, humerus, fibula, radius.)

Osteogenic sarcoma—The distribution of the combined Bristol Register and Cancer Campaign series of 140 osteogenic sarcomata *of all ages* is given in Table IX.

The general similarity of the anatomical distribution of these two tumours may be noted from Tables VIII and IX; but no sarcomata of the small tubular bones appeared in this series. Furthermore, it should be emphasised that osteogenic sarcoma of either pelvis or scapula is extremely rare before the age of fifty.

SEX DISTRIBUTION (of all ages and sites)

Comparable figures for the crude age and sex distribution of both tumours are given in Table X. The variations of the sex factors with age are shown graphically in Figure 12. It is

TABLE IX

OSTEOGENIC SARCOMA

Site	Type of bone	Number	Per cent	Combined per cent
Femur	Long	58	41·5	72·2
Tibia	Long	19	13·6	
Humerus	Long	17	12·2	
Fibula	Long	4	2·8	
Radius	Long	3	2·1	
Vertebra	Irregular	5	3·6	4·2
Tarsus	Irregular	1	0·6	
Scapula	Flat	4	2·9	17·9
Pelvis	Flat	18	12·9	
Ribs, sternum	Flat	3	2·1	
Skull	Membrane	8	5·7	5·7

of interest that these two curves are closely similar during the juvenile period, but not in the later decades.

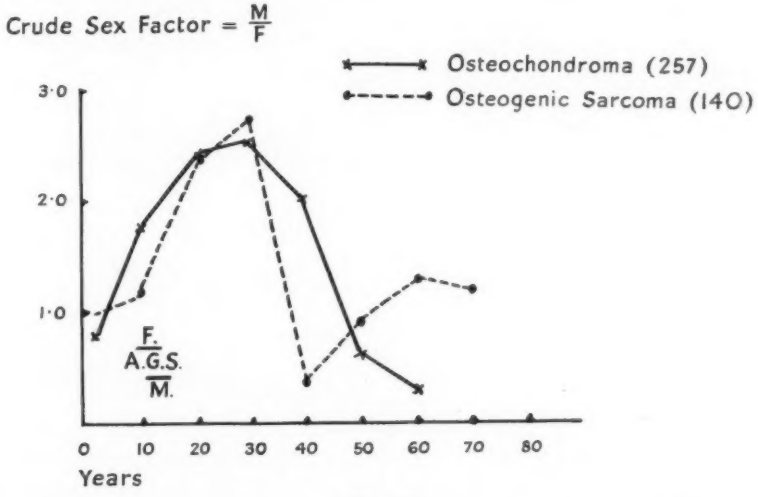

FIG. 12

Ratio of male to female (sex factor) in subjects of osteochondroma and osteogenic sarcoma at various ages up to thirty-five years. A.G.S.=Adolescent growth spurt.

RELATIONSHIP BETWEEN SEX AND SITE

Osteochondroma—The crude sex factor for tumours of the main bones in persons under thirty-five years of age is given in Table XI.

Reference to Table VI indicates the typical male preponderance for osteochondromata of the three major long bones—femur, tibia and humerus—which Table VII shows to obtain

in each decade. It is probably also present for the small tubular bones, metatarsals, metacarpals and phalanges.

Table VI, however, includes far too few tumours of flat bones to permit any useful conclusion about their sex distribution. However, by supplementing the data in Table VI

TABLE X

	0–	5–	15–	25–	35–	45–	55–	65–	Totals
*Osteochondroma**									
Males	4	63	45	27	18	5	1	—	163
Females	5	37	19	11	9	8	4	1	94
Sex factor—$\frac{M}{F}$	0·8	1·7	2·4	2·5	2·0	0·6	0·6	0·3	1·74‡
Osteogenic sarcoma†									
Males	1	13	19	8	2	7	14	16	80
Females	—	11	8	3	6	8	11	13	60
Sex factor—$\frac{M}{F}$	—	1·2	2·4	2·7	0·3	0·9	1·3	1·2	1·3

* Series from Geschickter and Copeland Tables 2 and 3 (1949).

† Combined Bristol B.T.R. and B.E.C.C. series.

‡ On pooling together the three large series of tumours in Table VIII (where information is available), the sex factor for all ages of 1·79 obtains—(538 M : 303 F).

with tumours of the scapula collected from other sources in this age group, a slight male predominance in numbers of tumours was found—M 16 : F 13. Stocks and Barrington (1925) recorded twenty-three scapular tumours of all ages—M 14 : F 9. Regarding osteochondromata of the pelvis, the same authors commented upon the marked reversal of the usual male

TABLE XI

Site	Type	Sex factor $\frac{M}{F}$	Source of data
Femur	Long	2·2	Table VI
Tibia	Long	3·3	Table VI
Humerus	Long	2·8	Table VI
Hand and foot	Tubular	1·6	Table VI
Scapula	Flat	1·2	*Vide infra*
Pelvis	Flat	0·2	*Vide infra*

preponderance, and recorded forty-one pelvic tumours in persons of all ages—M 8 : F 33. They further commented that this may well be due to the greater urgency of symptoms in the female, which would tend to be supported by the data here presented showing the relatively late age at which tumours appear at this site. Nevertheless, it should be pointed out that growth of the female pelvis is not only chronologically more advanced than in the male, but actually also greater in extent. This difference largely occurs at the time of the adolescent spurt in growth (Washburn 1948), with enlargement of the pubes and lower parts of the ilia. This is one of the more common tumour-bearing sites of the pelvis (Fig. 14), and even a relatively small mass here would probably call for removal on obstetric considerations.

Osteogenic sarcoma—Comparable figures for the relationship of sex and site for this tumour are given in Table V, the crude sex factors for the three major long bones being set out in Table XII.

It may be seen from Tables V and X that both with osteochondroma and osteogenic sarcoma the sex factor is considerably lower during the age period five to fourteen years than in

the next decade. In the latter tumour this is partly due to the relative frequency with which osteogenic sarcoma arises in the humerus in girls during this decennium. During the whole of the juvenile period sarcoma of flat and tubular bones is extremely rare.

TABLE XII

Site	Type	Sex factor $\frac{M}{F}$	Source of data
Femur . .	Long	1·6	Table V
Tibia . .	Long	1·9	
Humerus .	Long	1·0	

(All patients under thirty-five years of age.)

DISCUSSION

The data presented in the foregoing tables indicate certain similarities in the age, sex and site distribution of osteochondroma and osteogenic sarcoma, namely: 1) the total preponderance in males; 2) the earlier peak incidence and younger mean age of appearance of bone-forming tumours in the female; and 3) the anatomical distribution, and its variation with age and sex.

The male preponderance—This peculiarity appears over the main part of the life-span for both tumours. It is, however, less pronounced in early adolescence, and even reversed during some of the intermediate ages. The sex factor M/F is not of a high order, however; it is most evident in the " juvenile " patients here considered.

		0–34 years	All ages
Osteochondroma.	Sex factor M/F	2·3 (Table VI)	1·74 (Table X)
Osteogenic sarcoma.	Sex factor M/F	1·5 (Table V)	1·3 (Table X)

These factors may be compared with those for certain forms of soft-tissue sarcomata: lymphosarcoma 1·3 (Lilienfeld 1956); fibrosarcoma 0·94 (Stout 1948); fibrosarcoma 1·05 (British Empire Cancer Campaign 1949). Furthermore, it may be seen from Table X and Figure 12 that this sex differential is greatest between the ages of fifteen and thirty-four years. The features relating to age, sex and site distribution disclose the influence that skeletal growth has upon the presentation of bone-forming tumours, whether benign or malignant.

Stocks and Barrington (1925) showed the close correlation between the proportions of growth attributable to the two ends of the long bones and the number of osteochondromata that arise therein. If the proportions of growth in length are further corrected by a factor based upon relative cross-sectional area of the terminal growth cartilages, the agreement is even closer.

Anthropometric measurements show that the fully grown man is about 10 per cent bigger than the female in almost all respects. Thus the additional volume of endochondral bone formed during the rather longer growth period in boys may be roughly estimated to produce a correspondingly greater " risk " of tumour in the male. Since the main bone tumour-bearing period is related to the time of active skeletal growth, some allowance should also be made for its prolongation in the male. The factor fifteen-thirteenths may be applied if one assumes that bone growth in each sex is mainly completed by the end of the spurt at adolescence. The mathematical combination of these two factors of volume of tissue and time " at risk " may thus be given the values $1{\cdot}1^3 \times \frac{15}{13} = 1{\cdot}54$. This concept of the contributory factors that influence tumour causation would eliminate entirely the vexed suggestion formerly made of any greater

male liability to trauma. Theoretically also, the growth/size/time factors would be most strongly operative in the period when sex dimorphism is best shown; that is, after the completion of the adolescent spurt in both sexes.

The explanation here advanced to account for the greater male " risk " of bone neoplasia agrees with the experience of the experimental production of sarcoma by a variety of directly applied chemical agents, to which no increased susceptibility of male tissues has been demonstrated. Moreover, it is strongly supported by the anatomical distribution of the tumours listed in Tables VIII and IX, and in the distribution of sarcoma induced experimentally by the injection of widely acting agents such as radio-active phosphorus and strontium. In the dog, Cotchin (1956) stated that osteogenic sarcoma is not uncommon and is to be seen in the larger breeds (such as Great Danes), the bone most often involved being the humerus.

The more equal incidence in the two sexes (lower sex factors, M/F) seen in earlier years for both tumours (Table X) can be attributed to the relatively advanced female bone age at this time. Moreover, such information that has become available of the sex factor for tumours of scapula and pelvis, albeit inconclusive, broadly agrees with the characteristic sex differences in the growth of these two bones.

Multicentric or multiple forms of osteogenic sarcoma are very rare, being most often seen in older persons against a background of Paget's disease. Less than five acceptable cases of multiple osteogenic sarcomata have been reported in juveniles. Hereditary or familial forms of this neoplasm are likewise extremely few in the literature. Since with the innocent osteochondroma the male numerical preponderance is still higher among persons with a familial incidence (M/F=2·28, Stocks and Barrington 1925), it seems safer to accept the sex factor for the malignant tumour (1·5) as the best index denoting the actual effect of sex upon bone tumour incidence. In other words, the tumour distribution is random chance, subject only to features related to the sex dimorphism of skeletal development. One may add that male predominance of a like order is found in the condition " simple bone cyst "—a metaphysial disease that is degenerative and not neoplastic.

The earlier peak incidence of bone-forming tumours in the female—The female, even at birth, has a bone age a few weeks ahead of the male; the reason is unknown, but it has been well established that the average gestation period is slightly longer for girls. At the chronological age of ten years or so the difference has increased to months; and three years later, after the adolescent spurt, the sex difference in bone age is probably of the order of two years.

The characteristic juvenile peaks in incidence are not seen in the soft-tissue sarcomata mentioned above (Price 1955). There can be little doubt that the relatively earlier age at which both bone-forming tumours appear in girls as compared with boys is due to the advanced bone age of girls.

Osteochondroma—The advanced bone age of the girl—even during the middle of the first decade—is reflected in the relatively greater number of these tumours which appear in girls aged five to fourteen years. Underlying this tendency is the appearance of the adolescent spurt at age eleven years, as compared with thirteen years in boys. Furthermore, Tables VI and VII show that in the humerus of both sexes, in which bone growth in length is relatively advanced in early years, the tumours tend to arise at an earlier age than with the long bones of the leg. The difference in the commonest age of onset in the two sexes appears to apply also to tubular bones, but it is uncertain whether it obtains in the flat bones.

Osteogenic sarcoma—A similar explanation is advanced for the comparable differences in the distribution of the malignant tumours in relation to age and sex.

Anatomical (site) distribution, and its variation with age and sex—The site distribution of these bone-forming tumours of the skeleton supports the view that they stem from an aberration of the basic processes of bone growth in length and remodelling. They are much commoner at the sites where the greatest increase in length and size of the bone occurs. Confirmatory evidence of the importance of growth in the causation of juvenile tumours is supplied by

their well known predilection for the metaphysial region of the long bones. This feature is not nearly so clear in older persons, in whom the widespread damage of Paget's disease leads to more frequent involvement of the diaphysis. Of the thirty-six sarcomata considered in Tables II and III at least twenty-six (72 per cent) were metaphysial. Solitary osteochondroma also but rarely springs from the diaphysis of the long bone, indicating that the chondroid nodule from which it has grown was absent at birth, but has arisen after the host bone has grown in length.

In the flat bones (ilium and scapula) the common sites of origin of osteochondromata are depicted in Figures 13 and 14. For ease of description, the endochondral ossification of these two bones may be greatly simplified by regarding each as a narrow segment of a concave

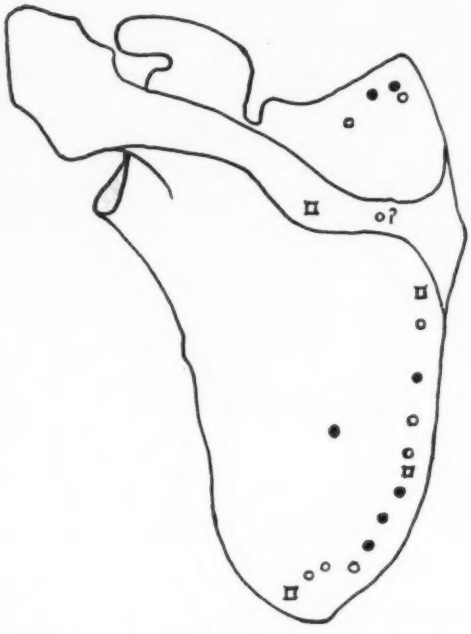

FIG. 13

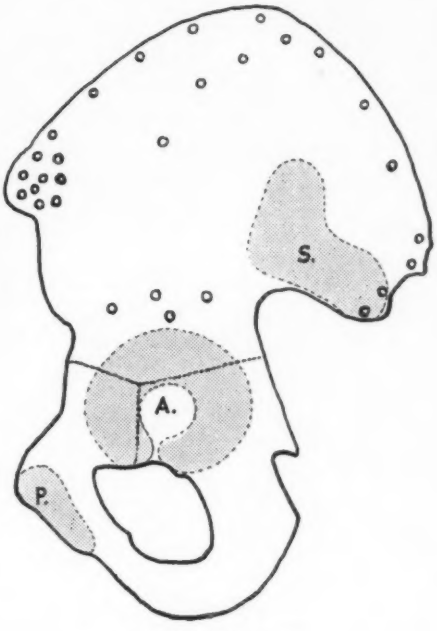

FIG. 14

Figure 13—Sites of twenty osteochondromata of the scapula. Mean age at onset 9·6 years. (Du Mortier *et al.* 15; B.B.T.R. 4; G. and C. 1.) Figure 14—Sites of twenty-nine osteochondromata of the ilium. Mean age at onset 20·6 years. (Ghormley *et al.* 25; B.B.T.R. 2; G. and C. 2.)

fan (Fig. 15). Osteochondromata usually arise in those parts of the flat bone most recently formed from cartilage, these areas being analogous to the more distinctive metaphyses of the appendicular long bones. During the juvenile age period defined in this paper, sarcoma of either scapula or ilium is extremely rare; it is only seen with any frequency in older persons usually having concomitant advanced Paget's disease.

The relationship between osteochondroma and osteogenic sarcoma—The foregoing analysis has indicated the close similarity of the age, sex and site distribution of osteochondroma and osteogenic sarcoma. From this, it may possibly be inferred that these two bone-forming tumours may have a closely related origin. The solitary osteochondroma rarely becomes malignant, and then most often in the form of chondrosarcoma derived from the growth cap. The frequency of this change has been given as 1·2 per cent of tumours by Stocks and Barrington (1925), under 2 per cent by Lichtenstein (1952) and 5 per cent by Geschickter

and Copeland (1949). Jaffe (1943) reported malignancy in 14 per cent of cases of a series of multiple osteochondromata; however, on considering this higher figure in the light of the number of tumours present in one person in the familial osteochondromatosis, there is still a relatively small " unit risk " of sarcoma.

Low-grade osteogenic sarcoma appears most frequently in adolescents or persons of early middle age. This tumour, in addition to relatively slow growth, shows good differentiation and a characteristic micro-anatomy. Even in this type, however, there is usually no indication of an origin from a pre-existing benign tumour. The micro-anatomy of osteochondroma shows a proliferating chondroid cap, ossification and resorptive zones, and periosteal bone

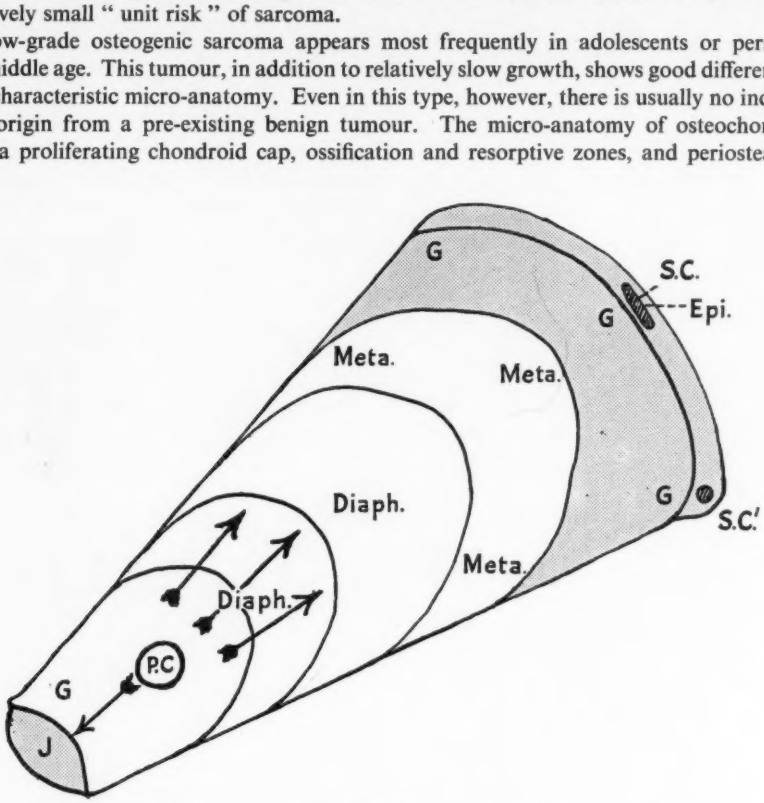

FIG. 15

Diagram representing ossification of " standard flat bone." " *Fan* " growth type. Relate to scapula or to ilium. P.C.—Primary centre of ossification, appears at second month (intra-uterine). S.C.—Secondary centre, appears in growth cartilage at fourteen to sixteen years. " Fan " growth stops about twenty-one to twenty-four years. S.C.[1]—Accessory secondary centre for angular prominence. This simplified concept of flat bone growth permits the identification of those parts of the bone which are analogous to the functional regions of a typical long bone: 1) Epiphysis (Epi.); 2) Growth cartilage (G.); 3) Metaphysis (Meta.); 4) Diaphysis (Diaph.).

formation in the pedicle—thus resembling a normal long bone (Fig. 2). These features in bizarre form may sometimes be observed in low-grade osteogenic sarcoma, but there is a subtle difference *ab initio* in the ability of the formative tumour cells to multiply indefinitely, to invade, and to destroy. Moreover, with increasing malignancy and more rapid growth, the sarcoma cells become more and more atypical.

Thus, although osteochondroma may potentially provide in its structure a suitable site for the appearance of sarcoma (as in any long-bone metaphysis), the great majority of osteogenic sarcomata take origin from tumour anlagen that differ initially from those that eventually develop into the benign osteochondromata. Nevertheless, both of these tumours in their observed form may show signs of reaction to the successive stimuli of normal physiological growth.

SUMMARY

1. Analyses are presented of comparable groups of the two commonest bone-forming tumours—osteochondroma and osteogenic sarcoma—derived from the following sources: Bristol Bone Tumour Register; British Empire Cancer Campaign Annual Report (1949); Stocks and Barrington (1925); Meyerding (1927); and Geschickter and Copeland (1949).

2. From this study, the following features emerge as characteristic and common to both tumours. 1) Both tumours are commoner in the male during the age periods 0–34 years, and over fifty years, with the possible exceptions of tumours of the humerus during early life and tumours of the pelvis. 2) The male preponderance is most marked for the appendicular long bones. 3) The male preponderance is greatest during the age period 15–34 years. 4) In the separate bones, these tumours tend to arise at a somewhat earlier age in the female. 5) In both sexes both tumours tend to arise at an earlier age in the bones of the upper arm and shoulder girdle than in those of the lower limb and pelvis. 6) Both tumours are related to an aberration of endochondral growth in length of bone.

3. The greater growth of the male, and differences in skeletal development in the two sexes, offer a simple and reasonably consistent explanation of these peculiarities and make it possible to discern the interplay of the three factors of age, sex and site of origin of the tumour.

4. From this concept it is possible to make a crude estimate of the factors of time and bulk of tissue which when combined may well account for the observed male preponderance of these tumours, and their anatomical distribution.

The author records his thanks to the contributory members of the Bristol Bone Tumour Registry, to many other surgeons, radiologists and pathologists who have referred cases, and to colleagues for their help in critical discussion, particularly Professor A. V. Neale for his advice and Mr G. M. Clarke for statistical assistance. The radiographs and photomicrographs are by Mr J. E. Hancock.

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IN MEMORIAM

ALFRED EDWARD WEBB-JOHNSON
1880–1958

His life was gentle; and the elements
So mix'd in him that Nature might stand up
And say to all the world "This was a man!"

The Right Honourable The Baron Webb-Johnson of Stoke-on-Trent

Knight Grand Cross of the Royal Victorian Order

Knight Bachelor and Councillor of the Imperial Society of Knights Bachelor

Commander of the Most Excellent Order of the British Empire

Hospitaller of the Grand Priory in the British Realm, Knight of Justice, and
Bailiff Grand Cross of the Most Venerable Order of the Hospital of St John of Jerusalem

Officer of the United Grand Lodge of Antient, Free and Accepted Masons of England

Doctor of Laws honoris causa of the University of Liverpool
and of the University of Toronto

Honorary Fellow of the Royal College of Surgeons of Edinburgh, in Ireland,
of Australia, of the Royal College of Physicians and Surgeons of Canada,
of the Royal Faculty of Physicians and Surgeons of Glasgow,
of the American College of Surgeons, of the Faculty of Anaesthetists and of
the Faculty of Dental Surgery in the Royal College of Surgeons of England

Honorary Fellow of the British Orthopaedic Association

Consultant Surgeon to the British Expeditionary Force—despatches thrice,
Distinguished Service Order, Chairman of the Army Medical Advisory Board

Lord High Steward of Newcastle-under-Lyme

Surgeon to Her Majesty Queen Mary

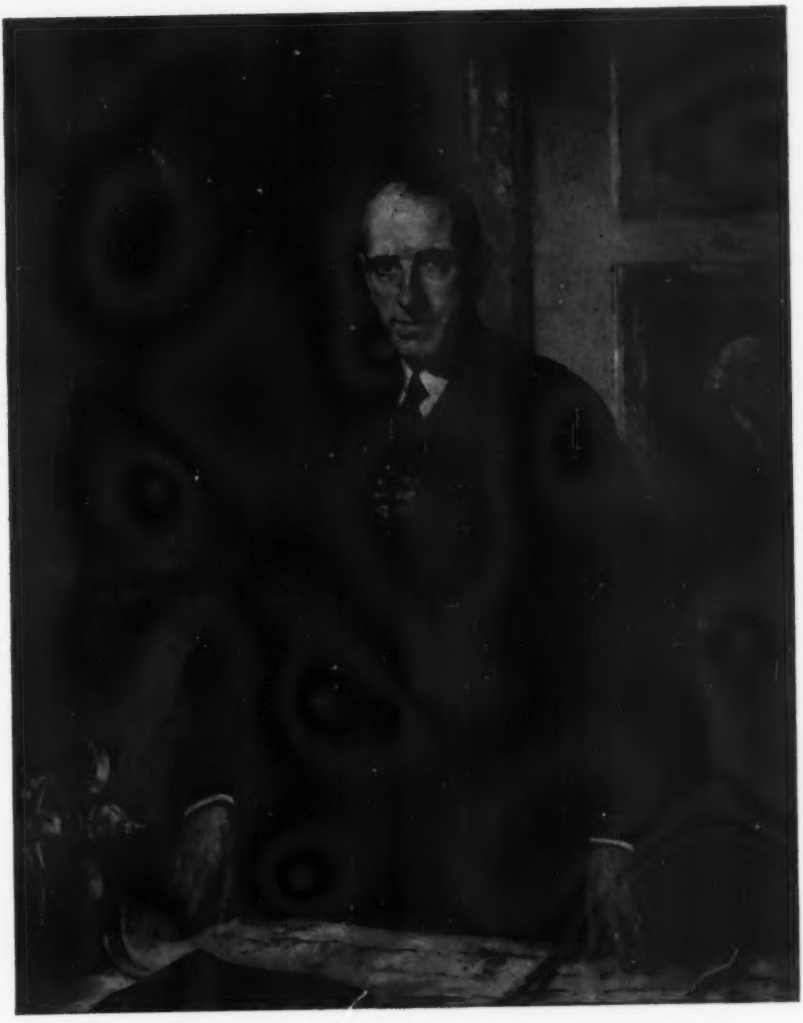
Dean of the Medical School, Consultant Surgeon and Vice-President of the Middlesex Hospital

President of Epsom College and President of the Royal Medical Benevolent Fund
President of the Section of History of Medicine and twice President of the
Royal Society of Medicine

Hunterian Professor, Bradshaw Lecturer, Member of the Court of Examiners,
Councillor, Member of the Court of Patrons, Honorary Gold Medallist,
and eight times President of the Royal College of Surgeons of England

For his width of vision and for the great contribution he made to his calling in his day and generation; for his long and successful service as President of the Royal College of Surgeons; for his drive and energy with such good humour that enabled him to succeed in great building schemes for the lasting benefit of Medical Science—especially remembering Middlesex Hospital, the Royal College of Surgeons and the Royal Society of Medicine; for his skill as a Surgeon which ranked very high; for his splendid influence on many generations of students; for his generosity in so many liberal acts, distinguished labours, researches and discoveries of the healing art—his life was a vocation. Many learned to follow his example with that same high purpose. For these and all the blessings of his life, we thank God.

From the Order of Service, St Margaret, Westminster, Thursday, June 26, 1958
Portrait by F. E. Hodge, R.B.A., from the Royal College of Surgeons of England



PROCEEDINGS AND REPORTS OF UNIVERSITIES COLLEGES, COUNCILS AND ASSOCIATIONS

UNITED STATES OF AMERICA

MEETING OF THE ORTHOPAEDIC ASSOCIATIONS OF THE ENGLISH-SPEAKING WORLD

ADDRESS BY THE PRESIDENT OF THE UNITED STATES

On May 14, 1958, the President of the United States addressed some of those attending the meeting at an informal gathering at the White House. President Eisenhower said:

Thank you very much. Needless to say, gentlemen, this is not any formal talk. It is my privilege, my honour, to greet you people here in the Nation's Capital and to felicitate you on the work you are doing in your professional capacity. But beyond such sentiments, I am concerned more with the residual good that comes out of a meeting such as yours—just because it is held, with all the English-speaking world here represented by men of education and of great and broad acquaintanceship, by reasons of their profession, among their own people.

It seems to me that this particular facet of a programme that I call—and we call here in this country—people-to-people, is a most important one and its results can be most fruitful.

I am convinced, from the top of my head to the bottom of my feet, that there is no enmity among people when those people get to know each other. The emotions, the uncontrolled prejudices and resentments that we sometimes encounter among people are normally because of the ignorance of those people as to the others—the people against whom they are resentful. The more we can spread the doctrine of people being guided by normal human aspirations, common objectives in life, common values that they treasure among themselves, by that much more we will advance the human race, not only in its joints and its bones but in its heart and its head and above all things in the way it can live in peace, and in opportunity of reaching—each individual and each race—its full capacities.

Now, on a more personal side, I hear that most of you people were involved in World War II, and as your chairman just pointed out, most of the men that were wounded in the chest and abdomen, they did not get into the medical places as often as they should have; but those that got it in the legs and arms and hips and bones, why there were lots of them. So you people obviously were very, very busy. And so it gives me a very personal reason for thanking, through you, all the orthopaedic surgeons of all the Allied Forces of World War II for the tremendous healing, the comfort and the help you brought to those men. Because only in your memories lives that knowledge of how many of them came into those hospitals, to defend the values of which I have just spoken—the priceless values of freedom and human dignity and the kind of thing for which all of us of the English-speaking world live.

So, to you each, good wishes while you are here, the hope that your meeting will not only be instructive but very enjoyable; that you will find the weather and other surroundings of the National Capital here the kind that will be conducive to your great pleasure.

And to all of the people with whom you meet back in your own countries, my warm personal greetings, and the wish that we could all meet together on this occasion to say "How do you do?" to each other. Goodbye, and good luck.

A TOUR OF ORTHOPAEDIC CENTRES IN THE EASTERN STATES AND IN CANADA

After the meeting, sixty-two of the visiting members embarked on a whirlwind tour of some eastern centres in the United States and in Canada, upon which the Washington standard of hospitality was maintained—it could not be excelled.

At Baltimore, in the New Auditorium of the Johns Hopkins Hospital, we were welcomed by Dr R. A. Robinson and entertained with a most varied programme of relevant papers by workers in various fields. *Dr G. Schultze, Dr E. Lang and Dr S. Eversole*, of the Departments of Radiology and Pathology, discussed disseminated lipogranulomatosis. *Dr M. M. Ravitch*, a general surgeon, described what could be done in case of chest deformity, especially funnel chest, which tends to be a much more serious disability than is generally supposed.

Dr W. M. Phelps, of the Children's Rehabilitation Institute, drew attention to the distinction of dystonia muscularis deformans progressiva from other causes of involuntary movement. Starting at

about six years of age with sprain of one ankle, for which support was not tolerated, spreading to the opposite hip and throughout lower limbs and trunk but rarely affecting the arms and never the cranial nerves or intelligence, the disease commonly progressed to death at about 15 or 16. Dr Phelps had observed arrest with large doses of vitamins B6 and E, and sometimes there was improvement with added quinine in doses preferably up to 30 gm. daily. *Dr J. P. Miller* demonstrated good correction of club hands, in which he had retained position by Kirschner wire transfixion for three months after operation. He thought five years the best age for this, younger patients doing less well. He also showed good results of the surgical treatment of unstable hips secondary to infantile pyarthrosis. Through an anterior incision he detached the gluteal muscles, performed a subtrochanteric adductor osteotomy of 130 degrees and inserted the greater trochanter into the acetabulum, reattaching the glutei more distally. A short plaster spica was worn for eight to twelve weeks. He had operated at $2\frac{1}{2}$, 3 and 13 years, but would like to do so at 6 to 7 months. In reviewing fractures about the hip, *Dr D. C. Finney, Dr R. B. Jones and Dr G. O. Eaton* confirmed current conclusions, including the high incidence of avascular necrosis in transcervical fractures. *Dr J. B. Brooks* described successful outcomes of laceration of the popliteal artery in femoral fracture.

In the afternoon of the same day we were able to see at the Alfred duPont Institute all that lavish philanthropy could bestow—" high heaven rejects the lore of nicely calculated less or more." Besides the hospital in its lovely setting we were privileged to see the grounds of Nemours, the seat of its fairy godmother, and to hear from our gracious host, Dr Al Shands, about the development of orthopaedic services, more particularly in the South.

A Sunday of fresh air at Atlantic City included a tour of the modern, beautifully designed and equipped Children's Seashore Home, under the direction of Dr Harvey N. Vandegrift. *Dr Thomas Gucker* reported a most interesting investigation of changes in respiratory function in scoliotics, whose vital capacity was often diminished when the spinal curvature was improved. Cases of atypical Legg-Calvé-Perthes' disease, acetabular protrusion and slipped upper femoral epiphysis, shown by *Dr Edgar L. Ralston and Dr Liebe Sokol*, led to lively discussion. *Dr Jesse T. Nicholson* showed cases of calcification at the elbow, fracture of the capitulum and pseudotumour in haemophilia, which led to a useful and comprehensive discussion of the modern treatment of haemophilic joints.

Taken by bus next morning to Philadelphia, we enjoyed further communications and discussions. Quite outstanding were cases demonstrated by *Dr Paul C. Colonna* in which he had carried out his operation of capsular arthroplasty for congenital dislocation of the hip. *Dr Z. B. Friedenberg* demonstrated benefit to badly paralysed patients from transfer of active biceps to triceps, enabling them to lift themselves from a chair; the ready dissociation from brachialis and hypertrophy of biceps were remarkable. *Dr J. R. Moore* showed some good results of his cartilage cup arthroplasty for non-united fractures of the neck of the femur, in which the greater trochanter capped by cartilage from the femoral head was thrust into the acetabulum. *Dr J. W. Lachman* shared the dissatisfaction of other surgeons with the results of treatment of ununited fractures of the lower end of the radius, and had fixed the lower ends of the radius and ulna with thin Stader pins, the free ends of which lay in stainless steel sleeves in the enveloping plaster. *Dr R. I. Peck* advocated the irradiation of osteosarcomas of the limbs before amputation. In discussing Apert's syndrome of acrocephaly with syndactyly, *Dr R. H. Cram* emphasised that the craniostenosis could be relieved by neurosurgery, granted early diagnosis. *Dr A. F. Seifer* drew attention to tibialis anterior as an obstacle in the reduction of Lisfranc's dislocation. *Dr J. T. Nicholson* described inclination of the glenoid in some cases of recurrent dislocation of the shoulder, *Dr E. L. Ralston* showed some cases of congenital vertical talus, *Dr M. S. Weider, Jun.*, of osteoid osteoma of the spine, and *Dr R. D. Heath* of myositis of the elbow without known injury and with a febrile onset, and *Dr H. H. Steel* discussed staphylococcus infections in hospital.

At New York, reached by bus the same day, we spent next morning at the New York Orthopaedic Hospital at the Presbyterian Medical Center under the chairmanship of Dr Frank E. Stinchfield and Dr Halford Hallet. *Dr C. Andrew L. Bassett and Dr James B. Campbell* gave an outstanding demonstration of the millipare tubulation technique for neural regeneration, in which nerve gaps were protected from centripetal fibroblastic ingrowth by tubes containing pores only one two-thousandth of an inch in diameter; encouraging neural regeneration had occurred. *Dr Walter A. L. Thompson and Dr Harvey Kopell* described a " pronator syndrome," in which disability in the pronated hand of a professional pianist was relieved on release of the median nerve from constriction between the ulnar head of the pronator teres and a band of flexor sublimis, happening only on pronation. *Dr Bradley L. Coley*, speaking from his immense experience, emphasised the fickleness of giant-cell tumours; and, with equal experience, *Dr Alan deForest Smith* gave considerations in the treatment of bone and joint tuberculosis, displaying unusual reluctance to carry out forage and the customary American readiness for arthrodesis. *Dr William H. von Lackum*, speaking of surgery in scoliosis, exhibited unusual optimism, supported by slides of good results. *Dr Charles S. Neer* showed some good results from shoulder prostheses in severe shoulder injury.

In the afternoon, at the Hospital for Joint Diseases, *Dr Leo Mayer* spoke of pelvic obliquity in poliomyelitis, to the treatment of which he has contributed so much, *Dr E. Kaplan* on tennis elbow and *Dr G. Selim* on osteomyelitis. *Dr Joseph Milgram* held a remarkable clinical discussion covering a wide range. Lateral tarsal pain after comminuted fractures of the calcaneum had been associated with encumbrance of the peronei, often by a boss of bone, which was more lastingly suppressed by the mallet than the osteotome. Function in arthrodesis of the shoulder had been greatly improved by excision of the clavicle lateral to the coraco-clavicular ligament. Among other interesting demonstrations Dr Milgram showed examples of the physiological reconstruction of tendon sheaths about temporarily inserted metal strips.

Inscribed silver bowl presented to the Washington Orthopaedic Club by the British Orthopaedic Association.

A morning was spent at the Hospital for Special Surgery, beautifully situated overlooking the East River at Cornell Medical Center. *Dr Philip D. Wilson*, Director of Research, welcomed us, and *Dr T. Campbell Thompson*, Chief of Surgery, presided. *Dr David M. Bosworth* of St Luke's Hospital spoke of tuberculosis of bones and joints in children, recalling the disastrous effect on the ear of prolonged administration of streptomycin, and discussed the place of Marsilid. *Dr P. Wade* gave a useful account of the operative treatment of ankle fractures, and *Dr Frederich R. Thompson* of the surgery of the osteoarthritic hip in which osteotomy received less attention than most of us accord it here. *Dr John R. Cobb* emphasised that in neurofibromatosis spinal curvature is short, sharp and angular, and may progress longer than other forms of scoliosis, so that watchfulness was necessary till the age of 21 at least. *Dr N. W. Littler* gave an outstanding demonstration of functional restoration after partial amputation of the hand, and *Dr Joseph E. Milgram*, of the Hospital for Joint Diseases, discussed in lively fashion the treatment of congenital dislocation of the hip, with special reference to the avoidance of vascular damage.

Next morning, at Toronto, a meeting was held at the Hospital for Sick Children, where *Dr F. P. Dewar* took the chair and we were welcomed by *Dr A. W. Farmer*, who showed a successful many-stage composite cross-leg graft for failed cases of tibial pseudarthrosis—an operation demanding all the resources of a master of orthopaedic and plastic surgery. By comparison, cases treated by the autogenous by-pass graft were shown by *Dr R. B. Salter*. *Dr W. T. Mustard* showed patients with outstandingly good results from ilio-psoas transfer to replace paralysed abductors—another operation that clearly needed the hand of a master and that recalled the great loss to orthopaedic surgery occasioned by the seduction of Dr Mustard's active and original mind and technical virtuosity to thoracic surgery. *Dr H. M. Coleman* showed admirable illustrations of his work on injuries of the

fibrocartilage of the wrist and the important part played by supination. A review by *Dr W. R. N. Lindsay* of Dr F. P. Dewar's cases of fractures of the calcaneum tended to confirm the virtue of conservatism. Some cases of radical excision of malignant tumours were shown: ischium by *Dr P. McGoey*, scapula by *Dr J. E. Hall* and humerus by *Dr D. L. MacIntosh*. *Dr W. R. Harris* showed recent advances in artificial limbs in the form of Canadian prostheses for Syme's and hip amputations. Dinner given us at the Hunt Club was the acme of happy conviviality.

Next day, at Montreal, we enjoyed an equally valuable series of demonstrations. *Dr Ted Shapter* and *Dr I. Bittenc* had calculated mathematically the pressure on the articular surfaces of the hip when taking body weight in various positions, which varied greatly and was least in abduction. They had consequently treated Legg-Calvé-Perthes' disease by allowing walking with the hips in plaster in this position. *Dr Roger Gariépy* discussed the treatment of rheumatoid arthritis of the knee by various methods, including high tibial osteotomy for flexion contracture. *Dr J.-C. Favreau* discussed limb equalisation, which he considered was called for with as little as one inch of inequality in the female and one and a half inches in the male. *Dr H. F. Moseley* showed the good results of various operations in severe shoulder conditions: excision of the head in fracture-dislocation, arthroplasty after such an injury, insertion of metallic prostheses in arrested caries sicca, repair of the musculo-tendinous cuff at various ages, and successful extra-articular reconstruction of the anterior glenoid lip in a severe epileptic. *Dr J. G. Shannon* spoke of his experiences in treating chronic osteomyelitis in collaboration with *Dr Woodburn* by means of immediate application of perforated split-skin grafts after saucerisation, with iliac chip grafting three months later. *Dr Languin* related the history of an inveterate bone cyst in a girl of 13 years. *Dr A. A. Butler* gave an interesting talk on scoliosis problems, especially correction and fixation in cases of extreme severity. Dinner given us at the University Club had the same happy touch that was so notable in Toronto, and we left feeling that Canada had something to teach us socially as well as academically.

A description of the final stage of the tour—at Boston—must await the next number.

WASHINGTON ORTHOPAEDIC CLUB

On the occasion of the Annual Banquet Mr Norman Capener, President of the British Orthopaedic Association, presented to Dr John P. Adams, President of the Washington Orthopaedic Club, a silver bowl of modern English craftsmanship as a token of gratitude for the generous hospitality extended by members of the Club to visiting British surgeons.

GREAT BRITAIN

BRITISH ORTHOPAEDIC ASSOCIATION

ELECTION OF FELLOWS, MEMBERS AND ASSOCIATES

At the Business Meeting in Washington on May 12, 1958, the following were elected Fellows:

E. W. O. Adkins (Derby)
J. Tulloch Brown (Motherwell)
L. M. Crooks (Aylesbury)
J. R. Hughes (Wrexham)

The following were elected Members:

C. G. Attenborough (London)
T. A. Berry (Durham)
P. W. Brand (Vellore)
A. Brown (Hove)
H. N. Burwell (Dewsbury)
C. S. Campbell (Bridge of Earn)
G. L. Clark (Bridge of Earn)
S. W. Davidson (Macclesfield)
H. L. Davies (Birkenhead)
H. Davis (Sutton)
M. B. Devas (Bexhill)
G. E. Dunkerley (Portsmouth)
G. I. Fenton (Londonderry)
A. W. Fowler (Bridgend)
T. B. Gardiner (Glasgow)
J. P. Heron (Liverpool)
J. Hirtenstein (Walsall)
W. Laurence (Hove)
T. E. McSweeney (Nantwich)
M. L. Mason (London)
J. D. Mineiro (Lisbon)
O. R. Nicholson (Auckland)
M. P. Pick (Dorchester)
C. M. C. Potter (Wolverhampton)
A. J. Sinclair (Doncaster)
C. C. Slack (Newcastle upon Tyne)
E. H. Strach (Liverpool)
P. R. Wheatley (London)

The following were elected Associates:

G. T. F. Braddock (London)
Douglas M. Caird (Perth)
J. R. Hochhauser (London)
Norman W. McLeod (Belfast)

J. Chalmers (Chicago)
P. F. Early (Manchester)
Peter A. Freeman (Glasgow)
L. C. L. Gonet (London)
B. J. S. Grogono (London)
Roland J. Hart (London)
Myer Makin (Jerusalem)
Ian K. Sharp (Hull)
E. L. Trickey (Stockport)
Samuel L. Weissman (Tel-Aviv)
V. H. Wheble (Northampton)

OXFORD UNIVERSITY AND REGION

During the last academic year the main activities of the Nuffield Orthopaedic Centre have been twofold. The clinical work in the wards, clinics and operating theatres has continued with the same intensity as in the preceding years. The number of attendances at the out-patient clinics, including both orthopaedic and accident cases, was over 50,000, and the number of operations for orthopaedic conditions, including trauma, was 2,850.

Teaching of the undergraduates of the Oxford Medical School, and training and research of postgraduates holding national, Dominions and foreign scholarships has followed the usual pattern in this centre. The Nuffield Scholar in Orthopaedic Surgery, Professor M. Makin from Jerusalem, has worked on osteogenesis in bladder mucosa transplants; and the Girdlestone Scholar, Dr B. Canfesci from Istanbul, is working on early calcification in the embryo. A number of other scholars have received, or still are receiving, postgraduate training or working in the laboratories. Dr T. J. Claffey, with a Scholarship from Sydney University, concluded his research on the vascular damage caused by several types of fracture of the femoral neck. A thesis on this work is being submitted to Sydney University. A Nuffield Dominions Clinical Assistant, Dr H. V. Crock, is studying methods of obtaining body-to-body spinal fusion. Dr W. M. Rigal, a Rhodes Scholar from Cape Town, is investigating osteogenesis in tissue culture. Dr G. Galli, from the University of Genoa, is finishing his work on the effect of tension across the epiphysial cartilage and upon growth. Dr A. Brodetti, from the Hospital for Special Surgery, New York, is studying methods of maintaining reduction in fractures of the femoral neck and their possible interference with the blood flow to the head. Seven other postgraduate students have been, or still are, carrying out clinical and experimental work.

During the last twelve months the Thursday evening lectures, initiated nearly ten years ago, have continued to be attended with as much interest as before. Among the lecturers must be mentioned in particular Professor C. Mensor from Stanford University, Dr J. Risser from Los Angeles, Professor A. K. Saha from Calcutta, Mr A. J. Helfet from the University of Cape Town, Dr J. D. Mineiro from Lisbon, Mr H. Jackson Burrows and Mr I. S. Smillie. There have also been various distinguished contributors from Oxford and its region.

The other aspect of the activities of the Centre during the present academic year concerns the erection of the new building where almost all the research and teaching functions will in the near future be concentrated. As was reported in this Journal (Vol. 38B) this splendid building has been made possible by the generosity of Lord Nuffield, who gave £200,000 for the purpose. The official opening of this building for research and teaching will be performed on October 27, 1958, by Her Majesty Queen Elizabeth the Queen Mother, who generously consented to enhance the ceremony with her presence as a token of appreciation both to Lord Nuffield and the Nuffield Orthopaedic Centre. A preview of the building will be possible for members of the British Orthopaedic Association during the autumn meeting to be held at the Nuffield Orthopaedic Centre from October 2-4.

The inauguration of the new building with its lecture hall for 150, library, museum, dining hall, canteen, common room, records office, out-patient and radiological departments, offices and laboratories, including those for electron microscopy, x-ray diffraction and biochemistry, will necessitate a change in the teaching policy of the Centre. It is likely that the Thursday evening lectures will be discontinued as a routine feature and that instead the first of the systematic courses will be organised.

The National Fund for Poliomyelitis Research has given over £35,000 to provide and organise an experimental Resettlement Centre under the direction of Professor J. Trueta in the grounds of the Nuffield Orthopaedic Centre, with the object of investigating suitable methods of completing the rehabilitation of severely paralysed patients by giving them specialised training in the use of devices, apparatus and gadgets which may make them as independent as possible. It is hoped that the unit will be functioning within a year.

THE HERITAGE, CHAILEY

MEMORIAL PLAQUE TO MURRAY LEVICK

On September 28, 1957, a plaque in memory of George Murray Levick was unveiled in one of the gymnasiums of the Heritage, Chailey. An associate of Sir Robert Jones, and an early Fellow of the British Orthopaedic Association, he was for many years Medical Director of the Heritage, which is

at once a boarding school on the lines of an English public school and a hospital for those children whose needs would not be met by a day school for the physically disabled, because their disabilities are too great or their homes too remote, or for other reasons.

Levick was a man of many parts. Qualified from St Bartholomew's and an officer of the Royal Navy, he was a valued member of Scott's last expedition, as Medical Officer and Zoologist. His powers of accurate observation produced his famous work on the *Social Habits of Antarctic Penguins*, criticised for its " anthropomorphic approach," and the standard *A Monograph of Adelie Penguins*.

After serving in the North Sea and at Gallipoli, where he was in the last party to leave, Levick became associated in physical medicine with Sir Robert Jones's work at Shepherd's Bush. Retiring from the Navy as Surgeon Commander, he became Consultant at St Thomas's and the Victoria Hospital for Children, and also Medical Director of the Heritage, which, under the influence of Robert Jones, was evolving from the old-style school for cripples into an active school and hospital with a full staff of consultants headed by R. C. Elmslie. Greatly impressed by the work of Rollier and Henry Gauvain, Levick became a practical advocate of open-air nursing, and established at the Heritage the necessary technique and trained staff for nursing children safely and happily out of doors, day and night, all the year round.

Small and wiry, Levick was apt at all games and enjoyed a discipline of physical fitness, but he never imposed these things upon others. In his younger days he founded the Royal Naval and Royal Marine Rugby Union. After the first world war he established the Public (later, British) Schools

GEORGE MURRAY LEVICK
1876—1956
Surgeon Commander, Royal Navy, 1902-17
Medical Director of The Heritage 1922-1950
Pioneer of Open air treatment
Polar explorer
Founder of the British Schools Exploring Society
A man of integrity, high purpose, simplicity and vision
A great leader

The Memorial Plaque

Exploring Society, of which he was Chief Leader. This Society, with the object of " developing self-reliance, endurance and fitness, and fostering the spirit of adventure in youth combined with a scientific training," conducted annual highly organised schoolboy explorations of uninhabited parts of Labrador Iceland, Lapland and elsewhere. This enterprise in itself was an achievement of the first magnitude.

Levick's knowledge and wise counsel were in wide demand; and, among his many other activities between the wars, he organised the training of blind masseurs in certain forms of electrical treatment, formerly altogether closed to them. In the second world war, at the age of 66, he was called from Naval retirement to train commandos.

It was characteristic that, when on his retirement from hospital work at 73, his colleagues gave him a dinner and a memento of their affection, he chose a new set of golf clubs.

Levick was the most steadfast, understanding, big-hearted and modest of men; and, perhaps because of his modesty, his great services never received the official recognition that they deserved. In 1942 he was awarded the Beck Grant by the Royal Geographical Society for services to exploration.

The plaque of unpolished lime, inscribed in Roman lettering and bearing insignia of the Royal Navy and of the British Schools Exploring Society, carved by Mr Joseph Cribb to the design of Mr Keir Hett, was unveiled by Sir Raymond Priestley, who spoke of Levick in terms of unreserved admiration and esteem. He knew him better than any other man, having spent, with four others, no less than nine months in an ice cave on Inexpressible Island when cut off by pack ice on Scott's expedition.

REGIONAL ORTHOPAEDIC CLUBS

SOUTH-EAST METROPOLITAN REGIONAL ORTHOPAEDIC CLUB

A meeting of the club was held at St Alfege's Hospital, Greenwich, on February 22, 1958. Clinical cases were presented by *Mr R. H. Sewell* and *Mr J. E. Buck*. These included the somewhat unusual condition of non-union of a Colles's fracture requiring bone grafting. There were also groups of hand injuries, knee injuries, a pair of "untreatable" club feet, and a remarkably interesting case of arthritis of the hips of obscure etiology. A demonstration of the use of the hot air bath in injuries of the hand was given by the physiotherapy staff.

Reconstruction of thumb—*Mr R. H. Sewell* had achieved a remarkable result by the use of an abdominal pedicle graft and later a bone graft from the iliac crest.

Varus hip—*Mr J. E. Buck* sought advice on the treatment of a case of varus hip in a boy aged 2 years, who had suffered neonatal osteomyelitis of the upper femoral metaphysis. Over a period of nine months the shortening of the limb had increased from an inch to an inch and a half, and radiographs indicated an increase in the deformity. *Mr H. L.-C. Wood* suggested that surgery should be avoided. *Mr R. C. F. Catterall* advised a late osteotomy, but *Mr J. H. Mayer* thought that an osteotomy should be done very soon. In view of this divergence of opinion Mr Buck asked for a vote and the majority was for delayed surgery—delayed as long as possible. A similar discussion followed on a case of pathological dislocation of the hip in a girl of 5, and here *Mr Wood* was alone in his wish to operate on the patient.

Club foot—*Mr Sewell* introduced the case of "untreatable" club feet in which the feet had been strapped within twenty-four hours of birth and had subsequently been repeatedly manipulated and later put on Denis Browne splints. The feet had remained very stiff, with marked adduction of the forefeet, and both tibiae had become progressively valgus in the lowest thirds. In the discussion *Mr A. C. Bingold* suggested that the tibial deformities were due to treatment fractures. Others gave various suggestions as to the priority of the tibial and foot deformities. Mr Sewell himself was in favour of soft-tissue correction as a temporising measure while the feet were growing, but most members thought that the longer surgery could be put off the simpler would be the final stabilising operations.

Unusual arthritis of both hips—*Mr Sewell* discussed a patient with bilateral arthritis of the hips in a man of 18. The condition had been present for just over two years and had originally appeared as a subacute arthritis of the left hip. Symptoms had been progressive and there had developed porosis of both hips and vertebrae, some intrapelvic protrusion of the hips and narrowing of the joint spaces. Histology of biopsy specimens from the capsule, synovium, bone and related lymph glands had been reported normal, as had extensive biochemical tests repeated after six months. Clinically there was now ankylosis of the left hip and much restriction of movement of the right. Suggested diagnoses varied from arachnodactyly to pre-psoriatic arthritis, and it was apparent that none of the speakers really knew the answer, but it was suggested that the Biochemical Unit at King's College Hospital might be able to throw further light on the problem.

Ligamentous injuries in the knee—*Mr Sewell* discussed ligamentous injuries to the knee with reference to the four cases he had shown. He advocated early repair and late weight bearing in isolated and some multiple ligamentous tears, and was supported by *Mr Catterall* and *Mr Mayer*. *Mr Buck* was a little cautious about surgery: he had recently suffered bilateral medial ligament sprains while ski-ing, and had devised a method of strapping whereby activity could be continued. In effect the method consisted of a strip of extension plaster applied with the knee and hip slightly flexed and the patient standing. The strapping passed from under the heel, outside the ankle, antero-medial to the knee and then upwards and outwards to end just above the iliac crest; this gave firm support to the damaged ligament and progress thereafter was almost painless.

Below-knee amputation for senile gangrene—*Mr Buck* described a case of senile gangrene in which a successful below-knee amputation had been carried out in a patient of 77. The patient was first seen with gangrene of two toes and slight inflammation of the whole dorsum of the foot. A mid-tarsal amputation had been performed but the foot flaps, though entirely unsutured, gradually began to slough, and a standard below-knee amputation was carried out, the flaps opposed only by a tulle-gras dressing. The main vessels were seen to be completely occluded at this level, and neither ligatures nor sutures of any kind were used throughout the operation. Sound healing had occurred in six weeks and the patient had been fitted for an artificial limb with ischial bearing.

Diabetic gangrene—*Mr Buck* also showed a case of diabetic gangrene in which a mid-metatarsal amputation had met with success, again without sutures or ligatures.

EAST ANGLIAN ORTHOPAEDIC CLUB

A meeting of the East Anglian Orthopaedic Club was held at the Royal Air Force Hospital, Ely, on January 25, 1958. The clinical demonstration was directed by Wing Commander D. M. Keir.

Osteomyelitis—*S/Ldr. J. G. Mander* showed six cases of osteomyelitis treated by antibiotics and drilling. The cavities were closed except for a small irrigation tube. The patients all did well but it was felt that the formation of a sequestrum in two cases was an indication that surgery had been delayed too long.

Diastematomyelia—*S/Ldr. K. R. J. De Belder* demonstrated a case of diastematomyelia. This was seen in a girl of 3½ who came complaining of weakness of her right leg, which tired quickly. Radiographs showed spina bifida in vertebrae T.4 and T.8 and an odd foramen was noticed in one of the bodies. The other signs noticed were fusion of some spinal bodies, with wedge-shaped vertebrae and fusion of adjacent laminae. S/Ldr. De Belder pointed out that a spicule of bone passed through a hole in the cord and that as growth progressed this exerted tension on the cord. The treatment consisted in removal of the spicule.

Divergence of middle and ring fingers—*S/Ldr. De Belder* showed a case of divergence of middle and ring fingers in a small boy. There was an ill defined lump between the metacarpal heads and café-au-lait patches on the skin. It was decided to explore the lump, probably a neurofibroma. No other lumps had been noticed.

Pes cavus—*S/Ldr. R. W. Povey* discussed a method of treatment of pes cavus and reviewed a number of cases. His method was to do a Steindler's division of the plantar fascia and to transplant the tendon of extensor hallucis longus to the metatarsal neck and the flexor tendons of the other toes to the extensor expansions. He also carried out interphalangeal fusion.

Osteochondritis dissecans—*W/Cdr. D. M. Keir* showed six examples of osteochondritis dissecans treated by open reduction and fixation of the fragment by one or more Smillie's nails. Post-operative treatment was by two weeks' immobilisation, followed by two weeks of weight-bearing with a guarding plaster. The nails were taken out six months later.

HOLLAND

SIXTIETH ANNIVERSARY OF DUTCH ORTHOPAEDIC SOCIETY

On May 2 and 3, 1958, the Dutch Orthopaedic Society celebrated its sixtieth anniversary. The meeting was held at the Nederlands Zeehospitium which is situated on the dunes just outside Scheveningen, a seaside resort near The Hague. The original hospital there was destroyed by the Germans but was rebuilt just over a year ago and opened by Queen Juliana in May 1957. The surgeon-in-charge is Dr de Mol van Otterloo, who was a most competent and genial host. The hospital has ninety beds, but it is hoped to add another sixty which will be devoted mainly to the treatment of rheumatic conditions. The Zeehospitium has a country hospital atmosphere about it and deals in the main with chronic conditions. It is characterised by the same cheery informality as one finds at Oswestry and Stanmore.

On the first morning the president of the Dutch Society, Dr G. F. J. M. Bär, gave an address on rehabilitation in relation to orthopaedics, which he thought should be the sole responsibility of the orthopaedic surgeon. After lunch on a pleasant veranda overlooking the sea three papers were presented. *Prof. P. Lacroix* (Louvain) reported some of his recent work on the healing of fractures; *Prof. W. Ehalt* (Graz) described the methods of rehabilitation used in Austria; and *Dr O. J. H. M. Verbeek* (Amsterdam) gave an encouraging account of his experiences with the use of homogenous grafts for the repair of ununited fractures.

In the evening there was a dinner at Scheveningen. There were a number of congratulatory speeches, unusual in being uniformly good; and what impressed the foreign visitors was the ability of their Dutch hosts to speak with equal felicity and wit in English, French or German as well as in their own language. The foreign guests were Dr J. Delchef and Prof. P. Lacroix (Belgium), Prof. W. Ehalt (Austria) and Sir Harry Platt and Mr H. J. Seddon. Mr Seddon presented the Dutch Association with a silver hammer, a gift from the British Orthopaedic Association to the senior Dutch Society. The " holly wood " required explanation.

The time of the meeting had been well chosen because on the Saturday morning the Society was able to visit the superb exhibition of bulbs at the Keukenhof. The Dutch bulb growers have recently acquired what must have been originally a most beautiful country park (complete with windmill) and now, each spring, they show how bulbs should look when planted out under proper conditions.

It was with some reluctance that the party returned to the hospital in the afternoon to resume its business. Mr Seddon gave an account of the methods he had found useful in the reconstructive surgery of the upper limb after poliomyelitis. Dr van Otterloo followed with a description of the practice of his hospital in the treatment of the convalescent stage in poliomyelitis, and the meeting concluded with a remarkable exhibition of films made by Prof. Ehalt which dealt with fractures of the calcaneum, and the treatment and rehabilitation of patients with traumatic paraplegia.

Silver-mounted hammer and block presented to the Dutch Orthopaedic Society by the British Orthopaedic Association. The base was made from holly wood grown in the grounds of the Princess Elizabeth Orthopaedic Hospital, Exeter.

ANNOUNCEMENTS

BRITISH ORTHOPAEDIC ASSOCIATION MEETINGS IN 1959

The Spring meeting will be held in Torquay and Exeter from April 30 to May 2, 1959. The French Society of Orthopaedics and Traumatology and the Belgian Orthopaedic Society have been invited to participate in this meeting. Those wishing to present a communication at the meeting should submit an abstract of about 400 words to the Honorary Secretary, British Orthopaedic Association, 47 Lincoln's Inn Fields, London, W.C.2, **at least three months** before the date of the meeting.

The Autumn meeting of the Association will be held in London in October 1959.

NATIONAL CONGRESS OF THE ITALIAN SOCIETY OF ORTHOPAEDICS AND TRAUMATOLOGY

The Forty-third Meeting of the Italian Society of Orthopaedics and Traumatology will be held in Padua under the presidency of Professor Calogero Casuccio on October 19–21, 1958.

The main subject for discussion will be " Bone grafts," especially the following aspects: generalities and biology of bone grafts (Dr Vigliani, Padova); autogenous bone grafts in man (Dr Stringa, Florence); homogenous bone grafts in man (Dr Mastragostino, Genoa); heterogenous bone grafts in man (Dr Agrifoglio, Genoa).

Orthopaedic surgeons from foreign countries wishing to participate should apply to: Segreteria del Congresso della Società Italiana di Ortopedia e Traumatologia, Clinica Ortopedica dell'Università di Padova.

Stress fractures—A Bureau of Stress Fractures is being formed at the Middlesex Hospital, London, England, by Mr M. B. Devas under the auspices of Mr Philip Wiles, for the purpose of further research.

Mr Devas asks that surgeons who have treated patients with this condition affecting any bone would be kind enough to send the details and the radiographs to him at the above address. He also hopes that patients might be referred for examination while still under treatment whenever this is possible.

Book Reviews

AN INTRODUCTION TO EXPERIMENTAL SURGICAL STUDIES. By W. J. DEMPSTER, F.R.C.S., Lecturer in Experimental Surgery, Postgraduate Medical School of London. 9×6 in. Pp. 463, with 72 figures. Index. 1957. Oxford: Blackwell Scientific Publications. Price 50s.

The appearance of this book is in keeping with modern trend and the awareness of the importance of this subject—experimental surgery. Although the author presents a strong and reasonable case for a full-time experimental surgeon, it must be appreciated that this field is open to and indeed necessary for all young surgeons. In obtaining their postgraduate qualifications they have studied again the basic sciences and, apart from the possibility of contributing to the advancement of their own speciality, they will develop from research practice a discipline of accurate recording, reasoning and self-criticism which will always be useful.

Many writers and speakers have considered the importance of research in orthopaedic surgery. McFarland (1952), writing on the clinical significance of experimental surgery, pointed out that many present day operations are " founded on neither experiment nor experience." Similarly, Harris (1948) has emphasised our responsibility of not depending upon others for acquiring knowledge of the " fundamental sciences " required in our work. Therefore, it is disappointing that in this book so little orthopaedic research is recorded, namely twenty-three pages out of 450 pages. This is not a criticism of its scope, but reflects how relatively little basic orthopaedic research is being carried out. However, in reading this book the orthopaedic surgeon will find much to interest him. Mr Dempster has achieved most admirably his aims of providing an excellent bibliography and critical commentary of a wide range of surgical research. In particular, the chapter on " Tissue Regeneration and Wound Healing," and that on " Tissue Grafting, Transplanting and Implanting," are singled out because of their most succinct and authoritative consideration and presentation. Other topics—such as low temperature studies, shock and cardiac arrest, anuria and fat embolism—are also clearly presented and will help the orthopaedic reader to appreciate some of the major advances in these fields.

In consequence of delay in publication, the bibliography covers the Western literature up to and including 1955. This makes us look forward to another edition with further commentary on other topics, including perhaps surgical studies in cancer research.

In conclusion, this important contribution to surgical literature is recommended to all, and it certainly should be available in all centres in which orthopaedic surgeons are being trained.—Robert B. DUTHIE.

HARRIS, R. I. (1948): The Future of Orthopaedic Surgery. Journal of Bone and Joint Surgery, **30-A**, 805.
MCFARLAND, B. (1954): Experimental Surgery. Journal of Bone and Joint Surgery, **36-B**, 527.

FUNCTIONAL BRACING OF THE UPPER EXTREMITIES. By Miles H. ANDERSON, Ed.D., Director, Prosthetics Education Project of Department of Surgery, School of Medicine, University of California, Los Angeles. Edited by Raymond E. SOLLARS, Associate Director, Prosthetics Education Project. 11½×8½ in. Pp. xv+463, with 457 pp. of figures. Index. 1958. Springfield, Illinois: Charles C. Thomas, Publisher. Oxford: Blackwell Scientific Publications. Price 72s.

This book is the first of its kind. In seven sections it covers the problem of paralysis of the upper limb as seen through the eyes of the instrument maker. The ingenuity of the reconstructive surgeon and his tendon transplants is matched by that of the brace-maker and his mechanical devices. Both strive to achieve functional independence for those who are severely paralysed. Some will consider that the splints recommended are too elaborate, and by adding weight to an already weakened limb may well defeat their own object: few will fail to be stimulated by the ingenious methods employed for activating paralysed limbs by remote control. Throughout the book it will be apparent that many of the ideas are adaptations of principles well recognised in the manufacture of limb prostheses. At the end of each descriptive chapter there follows a " do it yourself " section where the construction of splints is considered in great detail to the extent of listing the tools, equipment and materials

required for their manufacture. Illustrations are plentiful but their quality is not always good. A useful section on " gadgets " is included. Naturally, those interested in reconstructive surgery are chiefly concerned in ways of dispensing with apparatus. Nevertheless, there will always remain those patients in whom it is not possible to alleviate paralysis by surgery. It is particularly in these cases that the work of Dr Anderson and his collaborators can be applied effectively. The author is to be congratulated on providing a valuable reference book for all those concerned in the rehabilitation of the patient severely handicapped by poliomyelitis.—Donal BROOKS.

SURGERY IN INFANCY AND CHILDHOOD. A Handbook for Medical Students and General Practitioners. By Matthew WHITE, M.A., M.B., F.R.F.P.S. (Glas.), F.R.C.S. (Edin.), Consulting Surgeon, Royal Hospital for Sick Children, Glasgow; and Wallace M. DENNISON, M.D., F.R.F.P.S. (Glas.), F.R.C.S. (Edin.), F.I.C.S., Barclay Lecturer in Surgery in relation to Infancy and Childhood, University of Glasgow. Surgeon, Royal Hospital for Sick Children, Glasgow. 9×6 in. Pp. xii+444, with 266 figures, some in colour. Index. 1958. Edinburgh and London: E. & S. Livingstone Ltd. Price 45s.

This book by two distinguished paediatric surgeons of Glasgow offers the student of that school the theoretical knowledge that he requires to satisfy his examiners in the qualifying examination, which includes compulsory written and oral questions on paediatric surgery. In anticipation of this exercise the curriculum provides a ten-week course in this speciality during the final year and this book aims to supplement the clinical teaching which is available during this appointment. Orthopaedic surgery is inevitably an important part of the surgery of infancy and childhood, and consequently about one-third of this book is concerned with the disorders and injuries of the locomotor system. The authors have managed to compress this subject into about one hundred and fifty pages of text and illustration without omitting to introduce and define briefly any conditions commonly met in paediatric practice. This somewhat synoptic approach succeeds in making the book shorter to read and cheaper to buy but inevitably demands a certain dogmatism and perhaps oversimplification. The uncompromising statements that sternomastoid tumour is due to birth injury inflicted on a short muscle and arthrogryposis multiplex congenita to gross intra-uterine pressure are examples of some of the views expressed which might benefit from the qualification that a fuller treatment of the subject would allow. Within these limitations however the authors achieve the objective which they declare in their preface to have been their aim and the illustrations are of the high quality that seems inevitable when Livingstone of Edinburgh undertake a publication. By the same token however there is little in this book that will interest or provoke the orthopaedic surgeon—unless after reading those passages which concern his speciality he reflects upon the desirability of a division of orthopaedic surgery into two compartments the teaching and practice of which are distinguished one from the other by an arbitrary age barrier that anticipates by several years skeletal maturation.—G. C. LLOYD-ROBERTS.

HANDBUCH DER ORTHOPÄDIE. Herausgegeben von G. HOHMANN, emer. Direktor der Orthopädischen Universitäts-Poliklinik und Orthopädischen Klinik München; M. HACKENBROCH, Direktor der Orthopädischen Universitätsklinik Köln and K. LINDEMANN, Direktor der Orthopädischen Universitätsklinik Heidelberg-Schlierbach. In Vier Bänden, Band I Allgemeine Orthopädie. 10½×8 in. Pp. xix+1,186, with 632 figures. 1957. Stuttgart: Georg Thieme Verlag. Price DM 178.

This is a massive contribution edited by three of the outstanding German orthopaedists and written with the collaboration of some twenty authorities. In the introduction of this first volume the authors remind us that over fifty years ago the " magnum opus " in German first appeared in the contributions of Lorenz, Lange, Hoffa, Kölliker and other master surgeons of the time. This new book attempts to take the place of the old one as fifty years are too long a span for any continuity of thought to be possible in any intellectual sphere.

The effort and labour put into the collection of material is impressive, even if judging only by what has come out in this first volume of nearly 1,200 pages and 620 illustrations. This describes the pathology of the skeleton, muscles and nerves, and extends well into the generalisations of treatment after a chapter dedicated to the study of posture, mechanics of joint function and radiographic exposures.

As in all encyclopaedic books, though perhaps less in this than in others, there is some lack of balance between chapters, as, for instance, between that on degenerative arthritis, which is comprehensive (forty-two pages) and excellent, and that on acute haematogenous osteomyelitis, the whole problem being dismissed in about four pages. But in general the subjects are treated with a broad outlook which may be gathered by the rich and sometimes exhaustive bibliography at the end of each chapter.

This is a book that must take its proper place in the bookshelves of orthopaedic surgeons and which, I hope, one day will be published in English as there has been since the war too little information on the ideas and methods of our German colleagues.—Joseph TRUETA.

CHIRURGIE IN DER TÄGLICHEN PRAXIS. By Priv.-Doz. Dr. med. Henry NIGST, Chirurgische Universitätsklinik Basel. With Preface by Professor Dr. med. Rudolf Nissen, Direktor der Chirurgischen Universitätsklinik Basel. 10½×7½ in. Pp. 438, with 479 figures. Index. 1958. Stuttgart: Hippokrates-Verlag. Price DM 72.

Anyone who has attempted to compress an outline of general surgery into 400 pages will sympathise with Dr Nigst. According to the mood of the reviewer, he can either deplore the lamentable omissions or marvel at the inclusion of such a wide range of subjects. The standpoint chosen for this book is that of the general practitioner, and it gives a clear and well-illustrated account of most of the common surgical conditions that he might reasonably be called upon to treat. A very sharp line is drawn in the text between these cases and those requiring hospital admission or specialist advice. The book is clearly and stimulatingly written, and opens with a chapter on use of the new medicaments in surgical practice. The illustrations are excellent, often being given in series to make a line of treatment or a point absolutely clear. Although the practical aspects of all surgical treatment are touched on or collated, the distribution of the material and the choice of illustrations lead one to believe that the author is particularly interested in orthopaedics and traumatic surgery. There is an excellent set of photographs and accompanying text covering the treatment of the injured hand, with particular attention to skin loss.

Where space is so valuable, it seems hard to justify the use of a page for a comparative table of the rest periods given by various "classical" authors in Austria, America, England and France. Unexplained discrepancies confuse the issue. All one can deduce about union is that the Austrians are pessimistic, the Americans omniscient, the English uncertain and the French cynical.

It was a pleasure to find emphasised—both in text and picture—the need to rotate the ankle medially through 30 degrees if a satisfactory picture of the tibio-fibular syndosmosis is to be obtained. When the appreciation of a small point is carried as far as this, it is difficult to explain the inclusion of Figure 76 illustrating the different types of malleolar fracture, which, apart from inadequacy, shows combinations of bone and ligamentous injury that do not occur from indirect violence. These are small blemishes on an excellent work.

It is a melancholy thought that the conditions of service of many general practitioners in this country is such that they can hardly be expected to make the efforts in self-help that this happy monograph is designed to encourage.—J. G. BONNIN.

THE LIFE OF HUGH OWEN THOMAS. By David Le VAY, M.S., F.R.C.S., Orthopaedic Surgeon, S.E. Metropolitan Region. 9×6½ in. Pp. vii+135, with 14 plates and 13 figures (some in colour). 1956. Edinburgh and London: E. & S. Livingstone Ltd. Price 25s.

This is a book that revives timely interest in Hugh Owen Thomas on the eve of the centenary of his qualifying in medicine and of the birth of Robert Jones. The two played a prominent part in proclaiming the principles of orthopaedic surgery. Mr Le Vay has made a careful study of the ancestry of Thomas and has inquired about his forebears in the localities associated with the family in Anglesey and other parts of North Wales. It is a great advantage to anyone who wishes fully to understand the teaching of Thomas to know something about his immediate ancestors, of their skill in the handling of skeletal injuries, and of the peculiar knowledge they acquired and imparted to their descendants.

There has been considerable speculation about the country of origin of the first ancestral Thomas. About 1735 he and a younger brother were thrown up by the sea from a shipwreck, speaking an unfamiliar tongue which some thought was Spanish but might have been Manx or Gaelic for even a monoglot native of Anglesey would have understood a northern Celt as little as he would a stranger from Spain. H. O. Thomas himself believed his progenitor was of Spanish origin. The boy was adopted

by a childless couple named Thomas who farmed at Maes in north-west Anglesey; they called him Evan. He took a lively interest in the domestic animals and those of the countryside, acquiring some skill in treating their injuries. This experience led him to try his hand in the treatment of sprains and fractures of his neighbours. His fame as a bonesetter penetrated throughout North Wales but he remained at his farm, treating his neighbours without fee. He died in 1814 greatly lamented as a public benefactor; a memorial tablet to his memory was erected by Lord Bulkeley at the church of Llanfair (St Mary's). His son Richard ap Evan Thomas succeeded his father as a bonesetter. Following frequent custom he dropped "ap" and "Thomas" and was known as Richard Evans. He also remained at his farm, treating the injuries of his neighbours without fee. When he died in 1851 his cousin Ebenezer Thomas of Clynnog, a leading Welsh poet and literary critic, wrote a moving ode to his memory. Richard's eldest son, born in 1804, was named Evan Thomas after his grandfather. He differed from his father and grandfather in that he gave up farming and took up bonesetting as a whole-time calling. He migrated to Liverpool, then expanding rapidly as a great seaport, and opened his offices at 3 Great Crosshall Street in the centre of the seafaring community whom he treated with such success that he became the club doctor to societies of boiler makers and other bodies. He was anxious to co-operate with the medical profession and in the early days relations were cordial enough. But when his reputation penetrated beyond dockland and attracted patients of a different social order he began to meet hostility and even malice. Within a dozen years he had to face three actions in the law courts for malpraxis and was acquitted on each occasion, press and public strongly supporting him. These actions did the profession no good and only aroused in Evan Thomas a combative and aggressive spirit. He defended himself against every insult with vigour.

Evan Thomas had five sons, all of whom were sent to Edinburgh University and afterwards to University College, London, for medical training. Hugh Owen, the eldest, was born at Bodedern, Anglesey, in 1834. He was a small, thin and nervous boy. On one occasion he was injured under the left eye by a stone thrown by another lad; a painful ectropion ensued. In later years he wore a seaman's cap with peak tilted down over the irritable eye to protect it from cold winds and to screen the disfigurement. He qualified as a Member of the Royal College of Surgeons in 1857 and proceeded immediately for a short educational visit to Paris, bringing back some eye instruments which he copied and adapted to form tenotomes. He joined his father in practice at Great Crosshall Street for a year or so before starting practice on his own at 24 Hardy Street, but he moved in 1866 to greater accommodation at 11 Nelson Street. Soon he was obliged to build extra waiting and examination rooms, a photographic room and a workshop with a smith and leather worker. He converted 24 Hardy Street into a private hospital of eight beds with a trained nurse in charge. This was indeed an unparalleled establishment for a man who was ostensibly in general practice. It had in miniature the features of the future country orthopaedic hospital; it provided Robert Jones with a model in his pattern for Baschurch.

The next decade was an intensive experimental period spent in the study and treatment of bone and joint injury and disease. Mr Le Vay finds that Sydenham more than anyone else exercised the greatest influence on Thomas. He quotes many instances of their similar attitude to the natural history and cure of disease. Neither paid regard to authoritative opinion but each stressed the importance of careful clinical observation. Thomas noted with warm approval that Sydenham "advised to permit Nature to do her own work, requiring nothing of the physician but to regulate her when she is exorbitant and fortify her when she is weak."

Soon after Thomas qualified there was delivered at the Royal College of Surgeons a series of lectures by John Hilton on "Rest and Pain." They had a considerable influence on Thomas in his management of bone and joint disease; his guiding principle was the conviction that absolute rest was essential for repair of injured or diseased tissue. For several years he devoted his skill and ingenuity in devising splints to secure his ideal of rest for diseased joints. Many types were tried and discarded before the final design emerged, usually simple in appearance, free from vascular compression when in use, and cheap of production. His splints kept disordered joints immovable and at rest; thereby replacing the reflex protective spasm of muscles with its wasteful nervous energy. Once a splint was applied to the part he was unrelenting in the observance of the principle of enforced, uninterrupted and prolonged rest until the joint had recovered from disease. If a joint was distended with pus he carried out frequent aspiration but without disturbing its fixation by the splint. But he only kept his patient in bed until night pains and disturbed sleep had ceased. Once the joint was quiet he allowed the patient up on crutches wearing his splint and an iron patten on the shoe of the sound limb. It was a patient walking about with an unusual appliance that attracted the attention of Rushton Parker, a young Liverpool surgeon. This incident led to his meeting Hugh Owen Thomas and the formation of a life-long friendship. Parker urged Thomas to publish his teaching and splinting procedures. The result was the appearance in 1875 of *Diseases of the Hip, Knee and Ankle Joints.* This classic monograph is a veritable landmark in the history of orthopaedic surgery. He relates

his experience of the conservative treatment of joint disease of the lower limb. Numerous case histories are produced from which it is possible to trace the development of his ideas. The completely satisfactory result of his treatment of diseased joints by enforced, uninterrupted and prolonged rest secured by adequate splinting impelled Thomas to protest strongly against the prevailing practice of amputation or excision of joints with its high mortality. He complained that rest was not understood and that, when it was attempted, the way to secure it was so inefficient that it failed to cure; hence the hope falsely set on operation.

In the monograph the splints are depicted for the first time. Nothing is more symbolic of the genius of Hugh Owen Thomas than his knee splint. Introduced first for tuberculosis of the knee, it began as a splint of recumbency and later was found to permit treatment with walking; subsequently it was used for fractures of the lower limb, for which purpose it has proved peculiarly adaptable. Its astonishing use in two world wars is well known; the lives and limbs it saved were beyond reckoning and the suffering it prevented in transport was testimony to its efficiency. Simple though the splint may appear at a casual glance it conceals elements of design tested by trial and error. Such may be inferred by its original description by Thomas: " The upper crescent is formed of an iron ring $\frac{3}{8}$ths of an inch in thickness, varying according to the age and weight of the patient; the ring is nearly ovoid in shape and is covered with boiler felt and basil leather; from its upper and lower portions two iron rods pass down to the lower end of the machine, where may be noticed a small staple for retention purposes, only used for the reduction of flexion. The ovoid ring should join the inner stem, forming an angle of 55 degrees, which, when correctly padded, becomes reduced to 45 degrees. This arrangement of the splint will be the most acceptable for wearing . . . the anterior crescent (of the ovoid ring) is much straighter than the posterior one and that the inside stem is connected anteriorly to the centre of the nearly ovoid ring the (outer) stem being connected to the central and uppermost portion of the ovoid."

Having embarked upon publication Thomas continued at intervals to issue Contributions to Surgery and Medicine. Several monographs appeared embracing his teaching on diseases of joints, deformities, fractures and dislocations. He was particularly familiar with the natural history of tuberculous arthritis and much of his teaching on joint disease is derived from his experience in the management of this particular malady. He maintained that the main cause of ankylosis of an inflamed joint was friction and pressure between articular surfaces; immovable fixation avoided these evils. If gross tissue destruction was present ankylosis of varying degree usually occurred. Thomas often observed some hip flexion as an early sign of hip disease and he estimated the amount of flexion by a test long associated with his name. If the thigh on the normal side of a recumbent patient is fully flexed to abolish lumbar lordosis the amount of persisting flexion of the diseased hip is revealed.

With increasing experience Thomas evolved a reliable test of recovery. He regarded a joint cured of disease as " sound " and one with disease still active as " unsound." A sound joint was one whose muscles moved it without spasm and whose range of movement increased with use and without deformity. Conversely an unsound joint was one whose movements remained restricted and any attempt to increase their range evoked muscle spasm. The deformity of an unsound joint he straightened, with restoration of symmetry, by gradual corrective adaptation of the splint; for instance, abolition of hip flexion was achieved by first adapting the splint to the deformity, and straightening splint and limb a little at intervals of a few days.

In the treatment of fractures of long bones Thomas aimed at restoration of length and symmetry with the maintenance of perfect rest by splinting during consolidation whilst avoiding vascular compression. He was successful in obtaining union of long-standing ununited fractures when the fragments were in contact. This he achieved by repeated " percussion " with a rubber-capped mallet, followed by " damming " of the circulation by tying a piece of rubber tubing above and below the seat of fracture. In his later practice he came to depend more on damming than percussion. For fractures of the elbow the " gauge halter " was introduced, later modified as a collar and cuff. He was particularly successful in reducing long-standing dislocations of the shoulder, for which he used a special chair firmly fixed to the floor, with a steel ring to control the scapula whilst two or more assistants exerted steady traction on the limb.

In spite of his publications the teaching of Thomas failed to penetrate the profession. This was partly due to their production with paper covers at his own expense; and the narrative was not arranged as well as it might have been to attract the reader, although what he had to say in advancing his ideas was stated with clarity accompanied by good illustrative plates. There were some who disliked his teaching and practice and could not forget his ancestry. Thomas himself was often aggressive and little disposed to convince by persuasion. He kept aloof from scientific meetings where he could have proclaimed and discussed his teaching on the treatment of joint disease and injury.

But there were a discerning few in this country and America who appreciated the significance of his teaching, amongst whom was Edmund Owen, who did actually succeed in getting Thomas in

1887 to address the Harveian Society in London. In the United States Virgil P. Gibney of New York adopted the teaching and John Ridlon came to Liverpool to watch Thomas at work, returning home a convinced disciple and propagating the ideas of Thomas and demonstrating the use of the splints.

The teaching however did ultimately find general acceptance, and that in a way that Thomas hardly expected. In 1864, at the age of thirty, he married Elizabeth, daughter of Robert Jones of Rhyl; the marriage was completely happy. Mrs Thomas was a woman of profound Christian faith whose equanimity enabled her to revere her husband's character and surround his physical frailty with maternal tenderness. They had no children but they adopted a nephew of Mrs Thomas, a youth aged fifteen named Robert Jones. He became an apprentice to his uncle and later entered the Liverpool Medical School as a student. He qualified in 1878 and became an assistant to Thomas upon whose advice he later proceeded to the F.R.C.S., Edinburgh. In a few years he was appointed Honorary Surgeon to the Stanley Hospital and in 1889 to the Southern Hospital where he built up an orthopaedic clinic which became world famous.

Robert Jones had an exceptional orthopaedic training by Hugh Owen Thomas from his youth upwards, and by his hospital staff appointments he was able to extend the boundaries of orthopaedic surgery so that he became recognised as the greatest of orthopaedic surgeons. It is to Robert Jones that we owe the perpetuation of the teaching of Thomas and the introduction of the splints to the whole profession. During the first world war he became Inspector of Military Orthopaedics and toured the Western Front giving demonstrations at casualty clearing stations in the use of the Thomas splints and particularly the caliper, which became a permanent part of war equipment.

Hugh Owen Thomas worked unceasingly in his practice for over thirty years and never took a holiday. It is surprising how his frail body stood up to such pressure. He engaged in busy general practice, and through heredity and inclination became absorbed in the study and treatment of skeletal disease and injury. No other surgeon took care of so many fractures and dislocations in a single year. He devoted his nights to the designing of splints and the writing of contributions to medicine and surgery. He was an outstanding pioneer of orthopaedic surgery. Much of his teaching is now part of regular orthopaedic practice and has lost its identity, whilst some has been tempered through inevitable scientific discovery.

Thomas, worn out by overwork, died at fifty-seven years of age on January 11, 1891. He had given his life to the crippled poor by whom he was loved and respected. The widespread grief manifested amongst all classes at Liverpool by his passing was a moving spectacle; thousands attended his funeral.

Mr Le Vay's book has the merit of combining a biography of Hugh Owen Thomas together with his essential teaching gleaned from a careful perusal of his monographs. This book should make a particular appeal to younger orthopaedic surgeons to whom this remarkable man is but a legendary figure. The author, although captivated by the sheer genius of Thomas as an original thinker in surgery, yet maintains a balanced perspective by criticism of his blindness or obstinacy in opposing some procedures of proved value. Mr Le Vay has attempted an analysis of the character of Thomas in explanation of the polemical nature of his writings, of his comparative isolation and his neglect of contact with other surgeons. Part of this attitude is to be attributed to the pressure under which Thomas worked and his complete absorption in the welfare of his patients.

The Life of Hugh Owen Thomas is strongly recommended to all who wish for more intimate knowledge of the man himself and of the profound part he played in the evolution of orthopaedic surgery. The work has all the pleasing features associated with a publication by Livingstone.—A. Rocyn Jones.

SIR CHARLES BELL, His Life and Times. By Sir Gordon Gordon-Taylor, *K.B.E.*, *C.B.*, F.R.C.S., Honorary Consultant Surgeon to the Middlesex Hospital; and E. W. Walls, M.D., B.Sc., F.R.S. (Ed.), S.A. Courtauld Professor of Anatomy in the University of London at the Middlesex Hospital Medical School. $9 \times 6\frac{1}{2}$ in. Pp. xii+288, with 49 figures and frontispiece, four in colour. Index. 1958. Edinburgh and London: E. & S. Livingstone Ltd. Price 42s.

Sir Charles Bell, a Scot, was one of many who came south to find fame in London. Primarily he left his alma mater to gain an outlet for his great abilities which had been sown and nurtured but not fittingly accommodated in Edinburgh. This book is the story of his life as seen by two of his countrymen who have trodden much the same road and whose names, like Bell's, are closely linked with the knowledge of anatomy.

Bell quickly won a unique reputation as an anatomist, not only because of his skill in this subject, but for his teaching prowess and for his ability as a physiologist and as a draughtsman. He was a pioneer in surgical anatomy and when he joined the famous school in Windmill Street, founded some

sixty-six years before by William Hunter, it prospered under his care. Bell was an able surgeon with a mind for philosophy, with the touch, the observation and the reassurance of a clinician and an attractive flair for teaching. This combination endeared him to his students and brought credit to the Middlesex Hospital, to which he was appointed in 1824. That the authors' sole object is to raise Bell to the status of immaculate genius is not apparent, for they are careful to temper this ascendency with a touch of human failing.

As the story unfolds the reader is courageously taken aside to view for himself scenes of London a century and a half ago. A conducted tour through the anatomy schools of London includes not only an introduction to the famous anatomists of the day but also a peep at the body snatchers, the resurrectionists, the sack-'em-up men or what you will. There is a trip to the battlefield of Waterloo, hot on the heels of victory, where Bell found outlet for his surgical skill and humanity by working day and night on the wounded of both armies. Back in London one senses the eccentricity of the Georges, rubs shoulders with the regency bucks and press-gangs and is fortunate enough to meet many of the doyens of literature, art, music and drama and the leading men of politics, exploration and science. There is a vivid glance into the practice of surgery of the time and some thrilling moments are spent with Bell in the theatre, but inevitably one is led back to the sobriety of the dissecting room to witness the evolution of research into the structure and function of the nervous system, an endeavour which absorbed so much of his time and energy. The chapters dealing with this study are profound, because through Bell's research others were stimulated to enter the race to unravel the secrets of the nervous system and one is drawn inevitably into the acrimonious controversy as to who was the greatest genius of them all, Bell, Mayo or Majendie. Whatever his rightful due in this field, he emerges with at least the credit both of discovering the different functions of the spinal nerves and the creation of some immortal descriptions and paintings of the facial and other peripheral nerves. His achievements were in many spheres as he played the role of anatomist, research worker, surgeon, teacher, painter, philosopher and administrator, and in the final scene the authors leave no doubt of his greatness. "The place of Sir Charles Bell in the ranks of truly great ones of our profession is assured. Kindly, and in the best sense of the word, simple, he will be remembered as one who, loving his fellow men, laboured unceasingly in the relief of suffering; who, seeking to broaden man's comprehension of nature, met success with humility and adversity with fortitude; and in all things recognised a Power transcending that of the world."

His letters, addresses, scientific papers and descriptions of his paintings are listed in full in an extensive appendix. His introductory address on the opening of London University, discussing the difficulty of protecting the integrity of a university in a large metropolis, is a fine example of his worldly wisdom. Who better to have written this biography and given us the benefit of such able research, relevant knowledge and historical conception than Sir Gordon Gordon-Taylor and Professor E. W. Walls? This book is as delightfully written as it is pleasurable to read and an enjoyable reminder of some of the great characters of our British surgical heritage.—Philip NEWMAN.

RÖNTGENOLOGISCHE DIFFERENTIALDIAGNOSE DER KNOCHENERKRANKUNGEN. By Hans HELLNER and Hanno POPPE, Göttingen, assisted by Ilse LOHSTÖTER. 11½×9 in. Pp. viii+903, with 518 figures. Index. 1956. Stuttgart: Georg Thieme Verlag. Price DM 270.

This large volume considers the radiological differential diagnosis of bone disease. Of the 903 pages the greatest part is an atlas, arranged by anatomical areas, with profuse illustrations. Most of the radiographs are excellently reproduced, mainly in the negative. Each is accompanied by a brief case history and a discussion of the diagnosis. When generalised affections of the skeleton are present reference is made to illustrations of other parts of the skeleton elsewhere in the book. It is thus possible, when a film is difficult to interpret, to compare it with a number of lesions occurring at the same site. The remainder consists of a fuller discussion of differential diagnosis, a section of topographical charts of disease distribution, and an alphabetical encyclopaedia with very short summaries of the most important features and correlation of references in the text and illustrations.

Some of the material is unusual and excellent, but the English reader is liable to be confused by a considerable variation in nomenclature. The book is somewhat unbalanced. Tumours are represented in undue proportion, while the description and illustration of some well known conditions is scanty to the point of neglect. Scurvy, for instance, has no mention of the formation of subperiosteal haematomata. Only one osteoid osteoma is shown and this is, at least, atypical. Little reference is made to the work of the last ten years; and conditions like aneurysmal bone cysts, chondroblastoma and

traumatic cortical hyperostosis are ignored. Some of the diagnoses given are open to question and others appear frankly erroneous.

In a work of this size there should surely be space for a bibliography, but this has not been included. The book can be regarded as having only moderate value and should be used only by those who are reasonably well informed, because in places it is liable to be misleading. Even allowing for the great cost of producing a book of this nature the price is rather high.—Ronald O. MURRAY.

OUTLINE OF ORTHOPAEDICS. By John Crawford ADAMS, M.D., F.R.C.S., Orthopaedic Surgeon, St Mary's Hospital, London, and St Vincent's Hospital, Pinner; Assistant British Editor, *Journal of Bone and Joint Surgery*. Second edition. $8\frac{1}{2} \times 5\frac{1}{2}$ in. Pp. viii+428, with 301 figures. Index. 1958. Edinburgh and London: E. & S. Livingstone Ltd. Price 35s.

In reviewing the first edition of this book we predicted its success. This forecast has been fulfilled by the appearance of a second edition in the short space of two years. In the meantime many emendations have been made—most of them to keep the work abreast of the rapidly changing orthopaedic scene, but some to add point to an already concise and careful text. Nevertheless, this has been extended by three pages only.

The terminology of bone rarefaction has been made more precise, which is difficult when so much must be left unsaid. The very brief section on manipulation has been amplified, but remains unsatisfactory because it does not tell the student how to detect the cases that "cry out" for manipulation, which are not those of frankly stiff joints, though these head Mr Adams's list; a place is given, however, to manipulation under anaesthesia in the treatment of foot strain. Mr Adams has adjusted his topographical descriptions to current trends with admirable reserve, as witness: "Because of the risk of the later development of osteoarthritis [after osteochondritis dissecans of the knee] if a large part of the femoral condyle has been excavated some surgeons recommend that the loose fragment be replaced in position and fixed with a pin. It has been shown that the fragment may unite again with its bed, but it is by no means certain yet that the incidence of late osteoarthritis will be reduced by this method of treatment." Eosinophilic granuloma now finds a small place, and, with again proper caution, it is given as one only of the causes of Calvé's disease. Strains of apophysial insertions of tendons, at for instance the heel and the upper end of the tibia, have rightly been distinguished from "osteochondritis juvenilis." Avascular necrosis is recognised as a complication of slipping of the upper femoral epiphysis (or its treatment), and a radiograph of its fixation by a tri-pin nail has been replaced by one showing fixation with three narrow nails. This is characteristic of the thorough revision of the illustrations as well as the text. The outline of the treatment of congenital dislocation of the hip is brought up to date, and differences of practice are fairly stated. The section on cerebral palsy has been much improved, spastic and athetoid states being clearly distinguished; the important association with prematurity is not mentioned.

Clearly there will be frequent further editions.—H. Jackson BURROWS.

A TEXTBOOK OF FRACTURES AND RELATED INJURIES. By J. Grant BONNIN, M.B., B.S. (Melbourne), F.R.C.S. (England), Consultant Orthopaedic Surgeon, Central Middlesex Hospital; Late Orthopaedic Adviser, South-East Asia Command; Hunterian Professor, Royal College of Surgeons, etc. $9 \times 6\frac{1}{2}$ in. Pp. x+710, with 775 figures. Index. 1957. London: William Heinemann Medical Books Ltd. Price 84s.

This book is a larger edition of Mr Bonnin's work, *A Complete Outline of Fractures*, first published in 1941. The present volume follows, generally speaking, the text of the earlier editions though there have been some expansions and alterations in the light of present practice. It says a good deal for the soundness of the original work that there has been so little to alter.

This edition includes an excellent chapter on head injuries by Mr T. G. I. James, a section by Dr J. D. A. Gray on Chemotherapy and the use of Antibiotics, which is as up to date as anything in a text-book can be, and there is reproduced again the section on injuries of the face by Mr J. N. Barron; but the book is chiefly of value for Mr Bonnin's own work on the treatment of fractures.

As before, the first part of the book is a general study of fractures and their complications including shock, the treatment of wounds and infection, and the principles of treatment. The second half of the book deals with individual fractures. As in the previous editions, each section is clearly written with a summary of the relevant surgical anatomy and is profusely illustrated with many excellent diagrams and radiographs.

If one has to criticise the work, it is perhaps that more care has not been taken in revising and eliminating methods of treatment which are by now obsolete. Some of the illustrations for example, show treatment which is out of date; for instance, the Roger Anderson distraction machine on page 111 and similar distracting machines on page 103 must be very rarely used now, and one hopes that one would never see a patient with a fractured clavicle trussed up in such a horrible fashion as is illustrated on page 300 under the description of " Sayre's Method." In the text also there are one or two questionable statements. For example, it is surely unwise and even dangerous to state without qualification that where the radial pulse is absent in a patient with a supracondylar fracture, the brachial artery should be immediately explored. One raises these points because it would seem that a book of this kind is essentially designed for the young surgeon who may have to rely on it in an emergency when called upon to deal with fractures on his own responsibility, and it is for him that this book will be of great value. One may say, however, that these are isolated examples, and, in general, the text gives several alternative methods of treatment and reflects fairly accurately the practices in this country at present.

Production of the book is good; it is compact and easily handled, and one would have no hesitation in recommending it as a necessary text-book for the staff library of any hospital that has to deal with a large number of accidents.—A. J. WATSON.

THE CLOSED TREATMENT OF COMMON FRACTURES. By John CHARNLEY, B.Sc., M.B., F.R.C.S., Orthopaedic Surgeon, Manchester Royal Infirmary. Second edition. $10 \times 7\frac{1}{2}$ in. Pp. xii + 260, with 199 figures. Index. 1957. Edinburgh and London: E. & S. Livingstone Ltd. Price 50s.

Mr Charnley in the second edition of this work again succeeds in giving all his readers a very clear picture of the closed wound that is a fracture. In the preface he belittles the importance of this achievement. He should not be so modest.

The new edition has sixty-six additional pages and sixty-six more illustrations, and is 15/– more expensive than its predecessor. As a better, a wiser and a beautifully produced book, it is well worth the additional shillings. Its purpose is to advocate the closed treatment of fractures " on biological grounds." If the sole aim of fracture treatment is the achievement of bone union in the shortest time, very few " experienced fracture surgeons " will disagree with Mr Charnley's views. A certain amount of overlap of fracture ends often permits early and generous production of subperiosteal " woven bone " and subsequent sound cortical union, the acceptance of collapse of cancellous bone permits its close apposition that Mr Charnley rightly claims as essential to its union. When we strive for exact alignment of all fractures, either by closed or open methods, in order to achieve the return of completely normal function, we very often slow down the healing process. The first twenty-six pages in this book explain why this should be so.

Personally I see no great harm in open reduction if certain biological principles are respected. For example, Mr Charnley roundly condemns circumferential wiring; yet I always use this suture to obtain exact alignment of the fibular fracture in the common type of fracture-dislocation of the ankle. In this injury the periosteum is often widely stripped by the violence causing the injury, and a little more stripping, just sufficient to place the wire subperiosteally, permits an exact reduction both of the cortical and cancellous elements of the fracture. In several hundred such operations I have not yet observed the disaster Mr Charnley illustrates in Figure 21, page 19. If a circumferential wire is placed outside the periosteum or around bones with no intact periosteal hinge then this disaster may occur. Surely this is an example of the poor application of the suture, not of the method itself.

In the closed treatment of fractures certain biological principles must also be observed. Discussing greenstick fractures of the forearm in children, Mr Charnley advises that " the fracture should be deliberately completed to remove the ' spring action ' of the intact bridge which may induce the return of the original deformity." I assume that this is done by increasing the " backward angulation." Such a manipulation could place an excessive tension strain on the vessels in the flexor compartment of the forearm. This manipulation is unnecessary. The apparent " backward angulation " is in fact a supination deformity and can be corrected gently and quite securely by a single manipulation in full pronation. Again with the care of blood vessels in mind, I would be—as undoubtedly Mr Charnley

is—very careful in the manipulation illustrated on page 166. If this manipulation goes easily, then the nerves and vessels have ducked to one side; if they " forget to duck " they may be seriously damaged.

I have again enjoyed reading this book and recommend it to all interested in the treatment of fractures. Mr Charnley, in mentioning Arbuthnot Lane's *Operative Treatment of Fractures*, invites

FIG. 21
Destruction of bone by " encirclage " (*see text*).

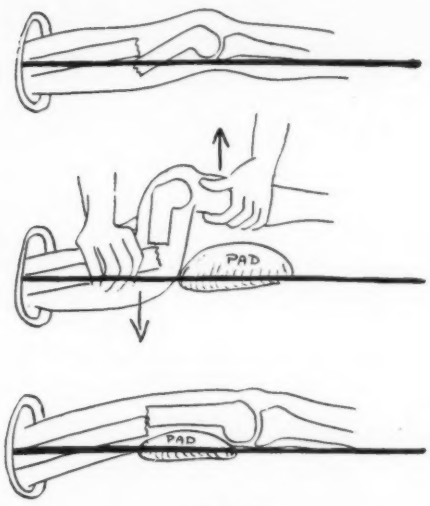


FIG. 132
Technique of reducing a transverse fracture in the lower third if longitudinal traction fails. Note that the master sling with its superimposed pad is applied to the splint *before* this manoeuvre is executed because it is an important fulcrum in the actual manipulation.

a comparison between the two methods. Undoubtedly the closed method of treatment of fractures is the safer method and results in a higher proportion of bone union. The functional results of the closed method, however, are not always satisfactory either for athletes or to meet the physical commitments of certain types of job. Yet if my practice in the treatment of fractures was restricted to one or other method then I would join the " Charnley Camp."—William GISSANE.

THE TREATMENT OF FRACTURES. By Lorenz BÖHLER, M.D., Director of the Accident Hospital, Vienna; Professor of Accident Surgery, University of Vienna. Volume III. Translated from the thirteenth German edition by Alfred WALLNER, M.D., and Otto RUSSE, M.D. 9½ × 7 in. Pp. xii + 803, with 932 figures. Index. 1958. New York and London: Grune & Stratton. Price £7, 7s.

This third volume has 803 pages of text. It deals with injuries of the knee joint and with all fractures, dislocations and sprains distal to the knee. It also includes several digressions, such as five pages on Painful Knees in Heavy Women and six on the position of the forefoot in flat foot, club foot and claw foot, illustrated by photographs taken in 1921. The last 137 pages are given to an appendix in which the author discusses the social, economic and other effects of injuries, the organisation of fracture-clinics and accident hospitals, disability evaluation, research in accident hospitals and, very briefly, his hopes for the future. This appendix is important, for it records the enormous influence of the Vienna Accident Hospital and of Professor Böhler himself. It reminds us that this book is the product of a great school and of a man who has made a lasting impact upon the treatment of the injured.

In discussing the injuries themselves this volume, like the earlier ones, repeats the Böhler principles and details the Böhler techniques. Acute knee injuries, ankle injuries and fractures in the leg and foot are classified very practically. There is an interesting brief review of the history of operations upon the fractured patella, but it overlooks E. S. Cooper's success in wiring that fracture sixteen years before Lister published his case. It is interesting that Professor Böhler is still faithful to wire sutures in this fracture, though he has " abandoned early operation [in closed fractures of the patella] because some failures followed inadequate operations by inexperienced surgeons " (page 1,523), surely an odd reason. It is interesting and strange that Professor Böhler sees rupture of the patellar tendon only in males, and stranger that this injury should have so relatively high an incidence in Austria among innkeepers and clergymen. Ruptures of the medial [tibial collateral] ligament of the knee are treated conservatively unless " the joint space can be opened to more than 15 millimetres " or unless the injury is more than ten days old. On the lateral side more laxity, up to 25 millimetres, is permitted within the limits of conservative treatment. Hey Groves's operation is advised for old ruptures of the cruciate ligaments.

Excision of the whole of a semilunar cartilage is condemned, as this will " usually cause later arthrotic changes " (page 1,609), which are prevented by leaving the rim of the meniscus behind. Intramedullary nailing is no longer advocated for fractures of the shafts of the leg bones, but the variety of techniques described includes closed intramedullary wiring. The screw-tractor is still advocated, and fluoroscopy is still used (Fig. 2411).

No one can accuse the author of neglecting to attend to detail. Plaster particles which have fallen into a case applied in treating closed fractures of the tibia and fibula " may be removed with a swab stick or a forceps, but a vacuum cleaner will do this much more quickly and better " (page 1,727).

Professor Böhler has found little reason to abandon the methods of the past, and no reason to omit their description. The Phelps-Gocht osteoclast, the calcaneal compression clamp and the screw-tractor are still advocated in certain heel fractures, though continuous traction on the toes has been given up in treating metatarsal fractures. Much use is made of transfixion wires in fractures of the bones of the foot.

It is impossible to avoid further reference to the " Questions We Should Ask Ourselves." As in the second volume, lists of these follow the descriptions of each injury and sometimes of individual techniques. Excluding occasions on which we are told that the relevant questions correspond to those appropriate to some other but related injury, there are forty-one such lists. The longest contains ninety-three questions to be asked " in Order to Avoid Failures in Fresh Open Fractures of the Tibia and Fibula," but the eighty-seven questions necessary in connection with " Pronation-Abduction Fractures of the Malleoli " run it close. This surgical soul-searching includes, in the case of the fractured patella, " Have I amputated at the proper time if infection has spread beyond the joint and generalised sepsis developed? " Less pessimistic inquisitions include, in the case of ankle fractures, " Have I asked the patient's name, age, occupation, body height and weight, and address? " Later, " Have I wrapped a paper bandage loosely over the cast so that the bed clothes are not stained by the ink? " On page 1,603 we are told that " diathermy, hot air and irradiation with lamps of different colours are time-filling but actually superfluous measures." Could not this admirable stricture have been applied equally to the writing of most of the " Questions We Must Ask Ourselves "?

There isn't much more to say. In reviewing the first two of these three volumes (Journal of Bone and Joint Surgery, 1957, **39-B,** 176, 608) I recorded my sorrow that an excellent practical book had swollen into a three-volume tome in which real wisdom was all but smothered in padding. Who was it who said that " The fate of books depends upon the capacity of the reader "? It is sad that this edition should tax it so much.—D. Ll. GRIFFITHS.